

**Experiences and perceptions of mothers of young people
with and without additional support needs and autistic
spectrum disorder in relation to behaviour, maternal stress,
access to services and family quality of life**

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ABSTRACT

This research explores experiences and perceptions of mothers of adolescents and young adults (13-22yrs) with Additional Support Needs (ASN) in relation to stress, service provision and family and individual Quality of Life (QoL). A particular focus is the effect of having a family member with ASN who also has a diagnosis of Autistic Spectrum Disorder (ASD) or where the young person with ASN may have an ASD which has not been diagnosed.

Mothers of young people from 4 groups were recruited: i) those with ASN and no ASD (n=41), ii) those with ASN and a diagnosis of ASD (n=18), iii) those with ASN, no diagnosis of ASD but with a positive score on an ASD screening measure (n=17) and iv) typically developing controls (n=17).

The mothers of young people with ASN (n=76) completed standardised questionnaires about family and individual QoL, stress, service provision, child behaviour and presence and severity of ASD traits. Twenty two of these mothers also took part in a semi-structured interview about coping with issues identified as most stressful by them in the stress questionnaire. The mothers of typically developing young people (n=17) completed standardised questionnaires on individual and family quality of life and on the behaviour of their similarly aged son or daughter.

Data collected via these questionnaires showed that increased severity of ASD was associated with increased maternal stress, which in turn was associated with decreased family and maternal QoL. Mothers of typically developing young people had significantly higher individual and family QoL scores than each of the three other groups.

The findings from the interviews supported the questionnaire results and gave further insight into mothers' life experiences. Mothers identified many perceived barriers to their child's progress including: lack of support and lack of co-ordinated service provision.

The results suggest that mothers of young people with ASN experience lower individual and family quality of life than mothers of typically developing young people. Parenting an adolescent or young adult with ASN is perceived as stressful and that the presence of behaviour associated with ASD is additionally stressful.

Possible reasons for differences in quality of life outcomes amongst the study groups are discussed. Implications for adequate services and recommendations for future research are suggested.

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Declaration

The participants who were the subject of this thesis were mothers of young people originally recruited into a larger parent study 'Cognitive impairment in schizophrenia: a clinical, imaging and genetic study of co-morbidity'. I was involved in recruitment and data collection for the parent study including administration of the CBCL and SCQ questionnaires which provided some of the data subsequently analysed and reported in this thesis. These data were also collected by other members of the research team, in particular, Jonathan Harris and Renate Kuenssberg, who also carried out the IQ testing in the young people.

This work has not been submitted for any other degree or professional qualification. I declare that this thesis is my own work, and the contribution to this thesis of others is clearly documented here and throughout the thesis where relevant.

Signed:_____

Dated:_____

CHAPTER ONE - INTRODUCTION

This research explores experiences and perceptions of mothers of adolescents and young adults (13-22yrs) with Additional Support Needs (ASN) in relation to stress, service provision and family and individual Quality of Life (QoL). A particular focus is the effect of having a family member with ASN who also has a diagnosis of Autistic Spectrum Disorder (ASD) or where the young person with ASN may have an ASD which has not been diagnosed.

This chapter describes how the thesis will be set out and explains the context of the research which was carried out as part of a larger research programme (the parent study). The parent study is described and how observations in the course of that work provided motivation for this research. In order to underpin clarity and economy in subsequent sections and chapters, operational definitions of the terms most frequently used throughout the thesis are given below. A full list of all abbreviations used in this thesis can be found in appendix 1.

This introductory chapter is followed in Chapter 2 by a review of the relevant literature. Chapter 3 presents the quantitative phase of the study, describing the methods and reporting, interpreting and discussing the results. Chapter 4 relates to the qualitative phase of the study, containing descriptions of the methods and discussion of findings. Chapter 5 draws together the findings of both studies and suggests areas for further investigation and implications for service delivery. Limitations of the study are discussed. The appendices contain a list of abbreviations with definitions, copies of data collection tools and all other documentation used in the study as well as lists of codes and themes emerging from the qualitative study.

1.1 Operational Definitions of Thesis Terms

1.1.1 Additional Support Needs (ASN)

The thesis refers to a group of young people described as having additional support needs (ASN). The term ASN is chosen as it is currently used within the education system from within which the sample was recruited. ASN refers to young people who have difficulties with education for a range of reasons, including intellectual disability (ID) (IQ< 70), autistic spectrum disorder (ASD) or any other factor causing a barrier to learning, such as social and emotional difficulties or attention deficit hyperactivity disorder (ADHD). The section on the parent study below (section 1.2.1) further describes the characteristics of the group from which the participants were recruited for this study.

1.1.2 Intellectual Disability (ID)

ID refers to people with IQ< 70, and this is a term interchangeable with learning disability in the UK and mental retardation in the United States. The IQ range 70-85 is defined as borderline ID and people in this group may also benefit from extra support in their education and in gaining independent living skills.

1.1.3 Autistic Spectrum Disorder (ASD)

In this study ASD is used as a broad definition to include all those on the autistic spectrum. The screening tool used in this study makes a distinction between those scoring in the autism range i.e. with a higher score than those in a middle range (who are on the spectrum but below the threshold for autism) and low scorers who are unaffected. Those scoring in the high range will be referred to as autistic and those in the middle range as having a pervasive developmental disorder (PDD); this may include those with a diagnosis of Asperger syndrome (AS).

1.1.4 Asperger Syndrome (AS)

AS is recognised as a condition within the autistic spectrum characterised by severe impairment in social interaction, but without the language delay or significant learning disability which often accompany autism.

1.2 The Parent Study

The research reported in this thesis was nested within a larger study, collecting additional data from families taking part in that study. The parent study was a large scale, MRC funded programme of research ‘Cognitive impairment in schizophrenia: a clinical, imaging and genetic study of co-morbidity’ aimed at determining whether it might be possible to identify young people who are likely to develop schizophrenic illness before they become unwell (Johnstone *et al* 2007). It was based on previous research showing that the prevalence rate of schizophrenia in those with a mild learning disability is at least three times that of the general population (Turner 1989) and on subsequent studies suggesting that cognitive changes may occur in the years prior to mental illness becoming apparent (Byrne *et al* 2003). The main hypothesis of the parent study is that amongst the population of young people with mild learning disability, it would be possible to predict, through structural magnetic resonance imaging (MRI), neuropsychological and genetic examination, those individuals likely to be later affected by schizophrenic illness. The ultimate aim of the study is to develop early intervention measures which may prevent or delay onset of illness and ameliorate symptoms when illness does occur. Although the parent study has a focus on schizophrenic illness, the young people recruited were not selected for any reason except that they had ASN; they were not in any other way considered to be at increased risk for mental health problems.

1.2.1 Recruitment to parent study

The sample of 501 young people was recruited through schools and colleges of further education (FE) across Scotland. The sample was recruited through education rather than health services to avoid the possibility of having an over representation of young people with a previous history of clinical input as this may not reflect the typical situation for young people with ASN; many such young people have very little contact with clinical services.

Local Education Authorities initially gave permission for the research team to approach Head Teachers of schools in their area. Head Teachers and learning support teachers in participating schools then identified the number of young people who met study criteria. In special schools teachers were asked to include pupils with mild to moderate learning disability, who had speech and did not have severe hearing or visual impairment, Down syndrome or known severe brain injury. Teachers in mainstream schools were asked to identify children with mild to moderate learning difficulties who required extensive support to access the mainstream curriculum for all or most of their school week.

The research team supplied the schools with the appropriate number of letters which the school then addressed and sent out to the parent with a covering letter. Each letter also contained a reply slip and a stamped addressed envelope and asked the parents to reply directly to the research team if they wished to participate. This ‘opt-in’ method allowed anonymity to be preserved until a parent chose to identify themselves as willing to take part by returning the consent slip.

In the case of further education colleges students were recruited from courses for students with learning difficulties. Students who were aged over sixteen were approached directly either by their teachers or were offered a letter of invitation during a class presentation from the research team. The letters could be passed on to a parent or other adult who knew them well if they wished to participate. As with the parent letters, participants were not identified to the research team until a slip was returned indicating that they were interested in taking part.

Although teachers in mainstream schools were asked to identify pupils who would be deemed to have a mild learning disability, because of the inclusive nature of education services in Scotland, the group of young people recruited were found on testing to have a broad range of IQ scores (40- 131). These young people had been identified by teachers as requiring learning support because of educational difficulties which appeared to be due to a range of conditions such as AS, ADHD, severe dyslexia or social, emotional and behavioural difficulties. These conditions had made learning in school difficult but did not always equate with a potential diagnosis of learning disability. The mean IQ of the 168 young people who subsequently became involved with the later stages of the parent project, tested using the WAIS/WISC (Wechsler 1981; Wechsler 1992), was found to be 73.

1.3 Motivation for this study

In the recruitment stages of the parent study, mothers of young people willing to participate were visited at home to complete questionnaires about their son's or daughter's personality and behaviour. During the course of discussions many mothers described aspects of their family situation and the challenges they had faced in relation to the care and education of their son or daughter. There was a recurring theme of dissatisfaction with service provision and support. The need for further investigation of the issues raised was apparent. This study was therefore designed to investigate the experiences of these families in a systematic way, aiming to identify factors contributing to increased stress and reduced family quality of life and also to identify unmet needs from the perspective of mothers, the main informants in the study.

The early screening for ASD in the parent study used the Social Communication Questionnaire (SCQ; Berument 1999). This showed that many young people scored in the PDD and autistic ranges although they did not have a diagnosis of ASD. Of those scoring above the threshold for ASD only 25% had a diagnosis. Since families often seemed dissatisfied with their access to information and support services the question was raised whether those families of young people screening positive for

ASD but without a diagnosis had had any particular difficulties compared with those scoring in the same range but with an ASD diagnosis as it seemed likely that those with undiagnosed ASD must not be receiving appropriate services and support.

A preliminary review of the literature showed evidence consistent with the reported experiences of families in the parent study and also documented some of the additional stresses which arise from having a family member with ASD. The literature also confirmed the possibility that many people with ASD within the community remain undiagnosed and that little is known about the implications of this situation for affected families. This study was therefore designed to explore the perceptions of mothers with reference to family quality of life, individual quality of life, stress and access to services and support. The study was also designed to capitalize on data collected in the parent study relating to child behaviour and autism screening data exploring these further in relation to the variables measured in this study. A qualitative phase to the research was also included in order to explore mothers' experiences in greater depth and to allow them the opportunity to voice their perceptions of unmet needs for their family.

CHAPTER TWO - REVIEW OF THE LITERATURE

In this chapter current and past research relevant to the focus of the thesis research will be described, showing the links between issues and highlighting key findings as well as gaps in knowledge. The four main areas of research reviewed are:

- 1) services and support for young people with ASN and their families
- 2) current debates in ASD
- 3) stress and coping in families of young people with ASN
- 4) conceptions of individual and family quality of life

The concluding section will draw together strands from each of the earlier sections to provide a rationale for the research questions and methods used in this study.

2.1 Services and support for young people with ASN and their families

This section will review research investigating services and support for mothers of young people with ASN as well as for the young people themselves and their families. In particular the current service provision in Scotland in terms of education, social care and health will be outlined and present inclusion policies will be described, including how these have changed in recent years. The implications of legislation such as the 'Education (Additional Support for Learning) (Scotland) Act' (Scottish Executive 2004) and the introduction of new initiatives based on recommendations from the Scottish Executive review of services 'The Same as You?' (Scottish Executive 2000a) such as co-ordinated support plans, person centred planning and local area coordinators will be explored.

2.1.1 Unmet needs for young people

Support needs for young people with ASN and their families will vary considerably according to different family situations and individual characteristics. In terms of the needs of young people themselves, marked difficulties have been described around adolescence, the period of transition to adulthood or to adult services (Freshwater and Leyden 1989; Stalker 2002). Those with mild ID have been shown to face challenges in terms of education, whether in special schools or within the mainstream. They are at risk of social isolation and bullying from their peers in the school environment as well as in their local communities (Stalker *et al* 1999; Cooney *et al* 2006). Many young people with mild ID or borderline ability ‘disappear’ from the system around the time they leave school (Caton and Kagan 2005). Suggested reasons for this include poor school attendance by these pupils and high levels of exclusions, difficulties in tracking post-school destinations and lack of alternative contacts once school links are severed. Caton and Kagan (2005) acknowledge that another reason may be the choice of individuals who no longer wish to be associated with the label of ID and deliberately break contact.

Problems of subsequent social isolation for young people and adults with ID have been reported in terms of loneliness and lack of friendships (McVilly *et al* 2006) and low levels of participation in socially inclusive leisure activities (Buttimer and Tierney 2005). Low levels of employment and associated barriers to people with ID gaining and sustaining employment have also been reported (McConkey and Mezza 2001; Rose *et al* 2005). It has been acknowledged that policies encouraging presence in the community are not necessarily able to address the degree of meaningful involvement and participation in community activities required to make a significant difference to the lives of people with ID (McConkey 1998; Cummins and Lau 2003) and as most people with ID live with their families (McConkey 2005) these issues affecting young adults with ID are likely to also have adverse effects on their mothers and families.

2.1.2 Unmet needs for mothers

McConkey (2005) reported a high level of unmet needs for carers of adults with ID in Northern Ireland: around 30% of carers had unmet needs for domiciliary support and around 32% had unmet needs for respite provision. A study in the Netherlands exploring support needs of parents of young people with moderate to borderline ID and emotional or behavioural problems also found a high degree of unmet need and reported a common perception of lack of information and counselling support (Douma *et al* 2006). Seeking support was related to parental perception of need rather than objective indicator of need and reasons for not seeking help included: not knowing where to look, problem not being serious enough or being temporary, and wanting to solve the problem themselves. Parents were more likely to seek informal support and only sought formal support when need increased. The most frequently reported needs were ‘a friendly ear’ and ‘information’; both of these provide emotional support for parents although information may also provide practical help in dealing with problems.

In a similar vein Hassall and colleagues (2005) found that while there was no association between level of service provision and maternal stress for mothers of children with ID, there was a significant association between helpfulness of service provision and family stress. The finding that the level of services does not have a significant effect on parents’ satisfaction suggests that the professional agenda in providing services may not be meeting parents’ needs. Parents have reported difficulties in dealing with the professional network and have highlighted the effort and work involved in negotiating service provision and in justifying their own and their child’s needs (Todd and Jones 2003; Beresford *et al* 2007).

Grant & Ramcharan (2001) identified a lack of research on family outcomes of service provision in the UK (particularly longitudinal studies); they also noted the absence of carers’ viewpoints and the perspective of people with ID in research to date. Douma *et al* (2006) also suggest that there is a lack of research on support needs of parents, particularly relating to co-ordinated care management and communication between parents and service providers. (Beresford *et al* 2007) found

that key outcomes for parents of disabled young people included maintaining an identity other than that of 'carer of disabled child' and related to this the need to be able to be a mother rather than a 'carer' for their child and to maintain family life for other children as well as the relationship with the other parent. The need for information and support to relieve stress was also recognised and was associated with financial resources and also the quality of the relationship with service providers.

An example of a mismatch between parental and professional perspectives could be the often cited goal of empowerment for parents whereby parents are encouraged to become advocates for their son or daughter. Nachshen and Jamieson (2000) in fact found that advocacy which was demanding in terms of time and effort or which did not have a successful outcome was perceived as an additional and unwelcome burden for parents.

Taken together these findings suggest that mothers perceive a significant need for informal emotional support and service provision of a high quality that is appropriate to their needs.

2.1.3 Unmet needs – ASD

Studies focusing on support needs of mothers with young people with ASD have reported similar findings to those given above. Bromley et al (2004) examined the impact of formal and informal support, unmet need and satisfaction with services on the psychological well-being of a group of 68 mothers of 71 children with autism. They found that most mothers reported unmet needs, particularly for practical support and respite care, and found that high levels of maternal psychological distress were associated with high levels of child behavioural problems and with low levels of informal family support. However, they did not find significant associations between the number of reported unmet needs and gender, severity of ASD or developmental delay, or access to formal supports. Wolf et al (1989) also found that maternal stress in mothers of autistic children was mediated by levels of social support although there was no such effect for fathers, suggesting a particular unmet need in mothers for informal social support.

Issues around transition for young people with ASD may be particularly difficult because of the inherent element of change. The Public Health Institute for Scotland (PHIS) produced a Needs Assessment Report in 2001 which set out a template for ideal service delivery. Needs for planned and well co-ordinated transitions were recognised, emphasising both the significant role of a named key worker or contact person to retain continuity and also the need for active involvement of the family and where possible the individual with ASD (PHIS 2001). Difficulties with sensitivity to noise, frequent changes of teacher and classroom and unsupportive peers with the subsequent risk of bullying and social exclusion may all make mainstream secondary school an uninviting prospect for a young person with ASD. Young people with AS, who are more likely to be placed in a mainstream educational setting, are reported as having particular difficulties with social interaction and also more susceptibility to behavioural and emotional disturbance than young people with high functioning autism (Tonge *et al* 1999).

The increase in prevalence of ASD, whether as a result of better ascertainment methods or a genuine increase, will also cause an increase in the number of families with substantial support needs (Fombonne *et al* 2001). High levels of maternal stress are likely to result in increased levels of need for affected families (Fombonne *et al* 2001). However, the expected prevalence is often not reflected in the numbers of people identified as having ASD (Brogan 2000). This suggests that there are likely to be many people with ASD who have not been diagnosed and this, along with the fact that service provision is often based on earlier estimates of prevalence, may result in a significant degree of unmet need for this population (PHIS 2001).

Another significant need for parents of children with ASD is for a sensitive and supportive professional approach to the manner in which the diagnosis is disclosed. Brogan & Knussen (2003) found that only 55% parents were satisfied with the disclosure of their child's ASD diagnosis. Factors contributing to satisfaction were: a definite diagnosis, a positive rating of the professional's manner, having the opportunity to ask questions and being provided with high quality information, including written information at time of diagnosis. This need for written information

for parents first receiving a diagnosis was recognised by a group of parents in Argyll in Scotland who campaigned and worked along with the Scottish Executive to produce an information pack to be made available to all parents at the time of diagnosis. This same group also worked on information resources for health professionals, in particular general practitioners as they may be the first point of contact for a concerned parent and yet may have had little training or experience in this area in terms of either young children, adolescents or adults with ASD (Scottish Executive 2006b).

This section has highlighted the reported unmet needs of young people with mild ID or ASN, including ASD, in relation to transition to adulthood, particularly noting difficulties relating to loss of contact with services and lack of social and employment opportunities. Unmet needs of parents have also been reported, such as their perceptions of lack of informal support although the link between the level of service provision and negative perceptions of parents has been questioned. These findings nevertheless suggest that the quality and appropriateness of services has an important influence on family outcomes and also highlight needs for psychological and emotional as well as practical support in terms of service provision. Many government policies related to service provision for children and adults with disabilities have changed over recent years and some of the relevant legislation and recommendations are described below.

2.1.4 Legislation and recommendations

There have been great changes over the last 40 years in the way society cares for and considers people with ID. In particular there have been many recent changes in Scotland in the provision of support to families of young people with ID. The population of people with ID is growing and is increasingly present in the mainstream of society, with most children and young adults with ID now living with their families (McConkey 2005). The importance of services and interventions aimed at the family rather than only towards the individual with ID has consequently been increasingly recognised (Scottish Executive 2000a; PHIS 2001) and embodied in a number of recent policy initiatives.

Children (Scotland) Act

The Children (Scotland) Act 1995 reflected these changes by emphasising the focus on rights and responsibilities of parents as well as requiring local authorities to provide appropriate services for 'children in need' which included children with disabilities. An aim of the act was to allow for more flexibility in providing support according to individual needs, while recognising the role of carers and families.

The Same as You?

In 2001 the Scottish Executive published a report entitled 'The Same as You? A Review of Services for People with a Learning Disability' (Scottish Executive 2000a); this report was followed by an English version called 'Valuing People' in 2001 (Department of Health 2001). The Scottish report 'The Same as You?' (SAY) informed a 10 year plan for the development of better services and support for people with ID in Scotland and made 29 wide-ranging recommendations, many of which are in the process of implementation. In particular the report attempted to ensure that all recommendations were acted upon by including a recommendation that each local authority would make a 'partnership in practice' (PiP) agreement about how they were going to implement the SAY recommendations in their authority. The Scottish Consortium on Learning Disability (SCLD) was set up in response to a SAY recommendation with a remit to advise and oversee implementation of the report recommendations and to ensure the participation of people with ID and their families and carers in the process. They also aim to provide training and information for professionals and to promote public awareness. They are currently carrying out research to report on the progress of implementation including reports from the perspective of service users (Curtice 2006).

Another recommendation of SAY was the introduction of Local Area Co-ordinators (LAC). This was based on an Australian scheme (Deschamp *et al* 2003) whereby a named professional in each local area supports a group of individuals or families and ensures that they have access to the services, support and funding that they need as well as building community resources for people with ID. The LAC would have a

network of links to services and knowledge of available supports and would be the first point of contact for families. This service has been introduced but provision varies across the country and by 2006 only 10% of people with ID in Scotland had access to a LAC (Scottish Executive 2006a). Other recommendations of SAY recognise the need to provide supports that are acceptable to service users and relevant to their needs, for example, the possibility of providing direct payments to service users and their carers wishing to manage their own budget in terms of service provision.

The SAY report also recommended that a network should be set up to deal with the needs of children and adults on the autistic spectrum. This was in recognition of the reported increased prevalence of ASD, and the lack of resources in education and support for employment to meet their needs and those of their families and carers. The Scottish Autism Service network, funded by the Scottish Executive, was set up in 2006 and works alongside the National Autistic Society (NAS) aiming to provide an information resource for people with ASD and their families and carers as well as a professional network to co-ordinate service provision. The NAS has similar aims on a national basis and actively campaigns for a commitment from statutory bodies and government agencies to increased investment in resources for people with ASD, including services, research and public awareness initiatives. In addition the NAS operate information centres for people with ASD, for carers and for professionals, they run volunteering networks to provide services such as befrienders and telephone help lines for advice and support (National Autistic Society 2006).

Health policy

Children and adults with ID are reported to be at increased risk of poor health care due to lack of appropriate health education, information and/or restricted ability to access services and also because of increased vulnerability to conditions such as epilepsy, mental health problems and certain physical health problems for particular groups (Emerson 2003b; Cooper *et al* 2006). In recognition of these difficulties, quality indicators have been developed by the National Health Service body NHS Quality Improvement Scotland (NHSQIS) and a programme of work monitoring the

progress of these indicators is on-going (NHS Quality Improvement Scotland 2006). The focus of these indicators is improving access to health care by including people with ID and their carers in developing and evaluating services. Improving health care for people with ID is also the focus of a report on the education of nurses entitled 'Promoting Health Supporting Inclusion' (Scottish Executive 2002). This report recommended that nurses training in all specialities should receive teaching to improve their awareness of ID and that people with ID and their carers should be involved in the development of the nurse education curriculum and the delivery of training, the latter being facilitated in collaboration with the SCLD.

Social Policy

The Social Policy Research Unit (SPRU) at the University of York (Beresford *et al* 2007) carried out a programme of research with disabled children their families and service providers in response to Government policy initiatives on child protection, looked after children and children at risk of social exclusion such as 'Every Child Matters' (Department for Education and Skills 2003). The SPRU set out to consider the implications of such policies from the perspective of disabled children and their families and to identify desired outcomes of service provision. They recognised firstly, that some of the policy framework outcomes were not appropriate or difficult to measure for children with various disabilities and secondly, that the additional stresses on parents of disabled children had not been fully addressed.

Key findings from this work showed that disabled children and young people wanted similar outcomes to their non-disabled peers, but that the issue of being able to communicate effectively and feeling safe needed particular attention for this group. Parents and carers identified the need for support to relieve stress, adequate information, financial resources and a positive relationship with service providers characterised by a sense of partnership.

Education policy

There have also been changes in education policy in relation to children and young people with ID. In 1999 the Riddell Committee was set up to investigate educational

and social development of children with severe or low incidence disabilities (Riddell Committee 1999). The report from this committee recommended a move towards mainstream education for all pupils who want it or whose parents want it. The practical application of this recommendation suggests that all children should first be considered for mainstream education with special provision being provided when the situation for the individual child does not meet a set of criteria. This was followed by legislation in the form of the Standards in Scotland's Schools Act (Scottish Executive 2000b) section 15 of this act set out the requirement of mainstream education which has become known as 'presumption of mainstreaming'. Other recommendations of the Riddell report included: more consultation with pupils and with parents about their wishes, better integration of services allowing access to fuller and more accurate information and also the setting up of an advisory forum to oversee developments.

The Beattie Committee was set up to identify the needs of young people with ID making the transition from school to further education, training or employment. This report too recommended policies of inclusiveness in further education and proposed that disadvantaged young people should be allocated a key worker to help them negotiate their educational choices with the different agencies involved (Beattie Committee 1999).

The Education (Additional Support for Learning) Scotland Act 2004 (Scottish Executive 2004) became law in Scotland in November 2005. The main changes brought about by this legislation related to empowering parents and young people and broadening the scope of support by introducing the term 'additional support needs' (ASN) to include all pupils who need additional support for any reason at any time in their school career. The Act requires all agencies (health, education, social work) to work together as necessary to support the young person. The previous 'record of needs' system was replaced, the new system requiring authorities to provide appropriate support to all pupils with ASN, although those with the most complex needs requiring help from different agencies may be eligible for a 'co-ordinated support plan', with this based around learning outcomes for the

child/young person. Provision is made for mediation, dispute resolution and tribunals for parents who are unhappy with the co-ordinated support plan. The stated aim of the Act is to develop a more personalised and goal-focused approach to education, with more direct involvement of parents and young people in decisions.

This inclusive approach to education encompasses the idea that children and young people with any level of ASN will be seen as part of the total population of school pupils and that provision should be made for the needs of all pupils within a mainstream setting. This replaces the more restricted view of integration as merely the policy of placing children with ASN in mainstream rather than special schools. Despite some resistance, evidence has suggested that the policy of inclusion has not had the detrimental effect, feared by some, on overall attainment of mainstream pupils (Dyson *et al* 2004; Pirrie *et al* 2006). However, although there now exists the ‘presumption of mainstreaming’ there is a consensus on the continuing need for a mix of provision, including special schools for children who require or desire their services. Special schools have not reported a significant change in the numbers of pupils on school rolls since mainstreaming policy was introduced (Pirrie *et al* 2006) suggesting that the demand for special provision has not declined in spite of policy changes.

Implications of changes

Over the last decade there have been changes in the perception of disability and moves towards greater inclusion in society for children and adults with ID. The main policy changes have been based on the recommendations of the SAY report in 2000 (Scottish Executive 2000a) and the Additional Support for Learning Act in 2004 (Scottish Executive 2004). However, research has suggested that problems remain in some areas, unmet needs reported for families include many issues which are part of the policy changes being implemented. This suggests that there may be a ‘theory-practice gap’, where research findings are not being acted on in such a way as to bring about real changes for those affected. New bodies such as the SCLD and the Scottish Autism Services Network have been set up and National Implementation

Groups are working on programmes of evaluation and implementation of SAY recommendations.

There are plans, by the SCLD, to introduce a database of people with learning disabilities in Scotland, the aim of this being to ensure sharing of information between agencies and to avoid the problem of service users having to repeat their story to each professional with whom they deal. A centralised database would also allow a review of services being provided and taken up by services users and may be a way of ensuring that people receive the support they need. A problem however with this kind of resource is that the numbers of potential service users are not known, this is particularly true for the group with mild learning disabilities as some in this group may not consider themselves as 'disabled' and may not come into contact with local authority services but only have their difficulties acknowledged in terms of their ASN. Although this group may not have marked impairments they still suffer disproportionately from the social isolation that comes from being perceived as being different, especially in the teenage years. Research also suggests that they are more likely to show challenging behaviour and to suffer from low self-esteem and mental health problems (Tonge *et al* 1999; Lecavalier *et al* 2006).

These findings suggest the need for proactive support for parents from service providers to prevent potential problems before they arise. It may be that parents do not want to see themselves as 'unable to cope' or 'needy' or 'pathological' and this may discourage them from seeking help. Counselling or support can help to raise self confidence and raise awareness and assistance in becoming involved with carer groups may help to provide peer support and access to information and services. The particular needs of single parents or those with financial difficulties can also be identified and measures put in place to address these needs.

2.1.5 Summary

There have been many changes over the last 40 years in societal attitudes towards ID and people with disabilities, with moves toward a more inclusive approach to education and to community provision of health and social services. New legislation and policies have supported this aim but may not have yet been successful in

providing opportunities for real inclusion in society. Young people with ASN are at risk of social exclusion and difficulties in finding employment, and suitable housing. Many young people with mild ID may be lost to the system after leaving school either by choice or through administrative failings, thus increasing vulnerability to negative future outcomes. Most young people with ID live with their families and mothers in particular have been shown to suffer increased stress due to a variety of causes. The level of formal support available may not be as important for maternal well-being as the perception of usefulness of support and as related informal emotional and psychological support.

Although most policies are now aimed at developing a co-ordinated approach to service provision and to improving communication, empowering parents and listening to young people, there is little evidence available as to whether or not family experiences reflect this change.

Questions arising from these findings include: What level of support do mothers report from various services and how useful is that support? What are mothers' perceptions of the challenges facing their own son or daughter and what resources are available to support them? Do the challenges and outcomes differ amongst mothers of those with a diagnosis of ASD and those with a high ASD screening score but no diagnosis and those without ASD? What changes would mothers like to see in service provision of their families?

2.2 Current debates in ASD

This section will describe the historical development of the concept of ASD and provide a definition of current understanding of the term. Debates on diagnosis, prevalence, causes and interventions reported within the literature will be discussed as well as suggested links to parental and sibling stress and family quality of life.

2.2.1 Definition

Autistic Spectrum Disorder (ASD) is a neurodevelopmental disorder, characterised by deficits in three main areas: social interaction, communication and imagination; this has come to be known as the triad of impairments (Wing and Gould 1979). Both of the two major main current diagnostic classifications of ASD, the Diagnostic and Statistical Manual of mental disorders, 4th revised edition (DSM-IV; American Psychiatric Association 1994) and the International Classification of Diseases, 10th revision (ICD-10; World Health Organisation 1992) are based on the presence of these three fundamental impairments. Although evident onset before the age of 3 years is requirement for a diagnosis of autism, the broader diagnosis of PDD can include those with a later age of onset as can the diagnosis of Asperger syndrome.

A definition by the Medical Research Council (MRC 2001) adds that ASD is a 'complex, debilitating and lifelong condition', emphasising the all-pervasive implications for those affected and challenging the misconception that autism is a condition restricted to childhood.

2.2.2 History

Autism was first described in 1943 by Austrian child psychiatrist Leo Kanner (Kanner 1943). His description of the characteristics and behaviour of a group of eleven children was the basis for what has become known as 'classic' or 'Kanner's' Autism. Hans Asperger, also an Austrian doctor and a contemporary of Kanner's, wrote a paper on autism in 1944, translated by Frith in 1991 (Asperger 1991). He described a similar group of children and also used the term 'autism'. Both Kanner and Asperger chose the same term to describe the characteristics of the groups they had studied, this was a co-incidence; the term 'autism' was not new at that time, having been first used by psychiatrist Eugene Bleuler in the early 20th century to describe a state of social withdrawal seen in association with schizophrenia (Bleuler 1916). While Kanner went on to gain universal recognition, Asperger was little heard of until after his death in 1980. In 1981 Asperger's work was reported by Wing (Wing 1981) who noted his descriptions of a group of more able children with

good language skills but with some of the same behavioural characteristics as the more severely affected autistic group and categorised this group using the term 'Asperger syndrome' (AS).

Early theorists described autism as 'childhood schizophrenia' or 'childhood psychosis' and the popular climate of psychodynamic theory at the time gave support to the theory that autism may have been the result of inadequate or abnormal parenting that had stifled the child's social and emotional development. Kanner himself gave some credence to this point of view although he had in his earliest writing described autism as having been present from before birth, recognising the likelihood of a biological rather than a psychopathological origin (Kanner 1943). During the 1960s and 1970s awareness of the condition increased and parents of children with autism became more vociferous, challenging the notion of poor parenting as a cause of autism. By the 1980s increasing scientific evidence, such as the higher rate of seizures in children with autism, suggested brain involvement (Volkmar and Nelson 1990) and twin and family studies showing the higher risk of ASD in siblings and particularly twins of affected children suggested a strong genetic component (Bailey *et al* 1995; Santangelo and Folstein 1999), resulting in an emerging consensus of autism as a neurodevelopmental disorder (Wing and Potter 2002).

2.2.3 Current explanations of autism

Happé (1994) suggested that autism can be understood on three different levels: biological, cognitive and behavioural, each contributing to our understanding of different aspects of the disorder. While the biological level is concerned with the genetic or neuroanatomical origins of ASD, the cognitive level seeks to understand the psychological mechanisms involved and the behavioural level focuses on the observed behavioural phenotype of those affected.

Three main neuropsychological models have been put forward to explain the deficits or cognitive characteristics of autism: theory of mind, central coherence and executive function. Each of these is briefly discussed below.

Theory of Mind

The deficit in theory of mind associated with autism has been described as ‘mind blindness’ or as a deficit in ‘mentalizing’ (Frith 2003) whereby an affected individual is not able to anticipate or imagine what another individual may think and thus to judge how they might react by being able to ‘put oneself in another’s place.’ This deficit in ASD has been demonstrated by a simple ‘false belief’ test where the individual is presented with a scenario involving two people and asked to make a judgement about the beliefs of one of the characters depicted. The test is based on whether the individual is able to identify the character’s false perception of the situation. Baron-Cohen et al (Baron-Cohen *et al* 1985) found that children with ASD performed significantly less well on this task than a matched group of children with Down Syndrome and deficits in this task have been reliably identified in many subsequent studies (Frith 2003).

Executive function

Executive function is the ability to think ahead and to plan actions with regard to consequences. Subjects with ASD showed impairments in this ability compared with controls in tests where they were required to deduce a rule for sorting cards (Bishop 1993) in another experiment Ozonoff et al (1994) also found that subjects with ASD performed less well than controls, matched for age and IQ, in a task where they were required to use forward planning skills to reproduce an arrangement of discs on pegs. Although difficulties in executive function are however by no means exclusive to ASD but occur in a variety of psychiatric disorders (Whyte *et al* 2006), the above studies found that deficits in executive function were significantly more widespread in people with ASD than in the general population.

Central coherence

Central coherence is the tendency to look for global meaning in what is perceived, for example when looking at a painting we may not recall all the details but will retain a general sense of what was being portrayed. The person affected by an ASD, due to weak central coherence, may not be able to grasp the global meaning but may concentrate on detail. This has been demonstrated by a superior performance in the

‘embedded figures test’ where the subject is required to identify a particular shape within a larger drawing (Shah and Frith 1983). It is this ability to focus on detail without being distracted by the whole that has been used to explain some of the ‘gifts’ commonly reported in autistic people or ‘savants’ such as remarkable memory skills or the ability to reproduce a detailed architectural drawing. Because of the potential benefits these abilities could bring, having ‘weak central coherence’ has been described as an alternative cognitive style rather than a deficit (Happé 1994).

These 3 neuropsychological models help to explain the social impairments related to ASD, however, another difficulty in defining the disorder is the great range of ability in those affected as well as differences in the degree of deficit experienced. This has led to a range of disorders being identified within a broader spectrum with the resulting diagnostic category of ASD.

2.2.4 Autism as a spectrum disorder

Since AS was first described (Wing 1981) there has been debate about whether it is part of the same syndrome as autism and in particular, whether there is a real distinction between high functioning autism and AS (Tonge *et al* 1999; Ghaziuddin and Mountain-Kimchi 2004). One possible answer to this question is to consider the condition as a spectrum disorder that can be manifest to different degrees in different individuals but with a common set of underlying features: the triad of impairments in social interaction, communication and imagination or a narrow range of interests or activities (Wing and Gould 1979). A reflection of the acceptance of the concept of a spectrum disorder was the inclusion of the category Pervasive Developmental Disorder (PDD) in DSM III (American Psychiatric Association 1980) with the addition of sub-groups ‘autistic disorder’ and ‘pervasive developmental disorder not otherwise specified’ (PDD-NOS) in the DSM-III-Revised (American Psychiatric Association 1987). In the current version DSM-IV (American Psychiatric Association 1994), new subgroups have been added with similar groupings being recognised by ICD 10 (World Health Organisation 1992). The current diagnostic criteria retain an overall category of PDD, with sub groups of: autistic disorder,

Asperger's disorder, PDD-NOS, Rett's disorder and childhood disintegrative disorder.

2.2.5 Prevalence

Autism was initially considered to be an extremely rare condition with an estimated prevalence of 2-4 per 10,000 children (Lotter 1966). However, reviews of prevalence studies since the 1960s have shown that although variation in reported prevalence may be due to differing diagnostic criteria being used or populations being studied, there has nevertheless been a significant increase in cases over time (Fombonne 1999; Wing and Potter 2002). A PDD prevalence of 26.1 per 10,000 was found in a British study of child mental health (Fombonne *et al* 2001) although prevalence rates as high as 58 per 10,000 have also been reported (Ehlers and Gillberg 1993). The MRC review of autism research (2001) estimated that approximately 60 per 10,000 children are affected by ASD and that 10-30 of these cases would be narrowly defined autism. The National Autistic Society estimated the prevalence of ASD as even higher, at 91 per 10,000. It was suggested that this would be made up of 20% individuals with autism, 36% with AS and a further 35% who do not quite fit the diagnostic categorisation of autism. However, those in the third category would be likely to be high functioning, undiagnosed and possibly not requiring any additional services or support. This led to a final estimated prevalence of 56 per 10,000 of individuals with ASD requiring some degree of specialist support (National Autistic Society 1997), very much in line with the MRC current estimate.

There has been considerable controversy as to whether there has been a genuine increase or whether the reported increase has been due to such factors as improved diagnostic methods and changes in perception of the disorder (MRC 2001; Wing and Potter 2002). These changes included the widening of the criteria after the recognition of AS in the 1980s and the subsequent development of the notion of a spectrum disorder. A true rise in prevalence cannot be ruled out however and suggested reasons for this have included: environmental factors such as viral infection, dietary minerals, vaccines (e.g. MMR) or exposure to mercury (used as a preservative in vaccines, although not in MMR). However, available evidence does

not support any of these causes (Taylor *et al* 1999; Fombonne and Chakrabarti 2001; MRC 2001).

2.2.6 Causes of ASD

Although ASD is now a well recognised condition, there are a wide range of manifestations of the disorder and many researchers have concluded that there are a variety of causes although the strongest influence is likely to be genetic, with a variety of genes implicated and with the possibility of a genetic susceptibility being triggered by exposure to an environmental risk factor (MRC 2001; MRC 2002; Volkmar and Pauls 2003, Happe, 2006). Links with genetic conditions such as fragile X syndrome (Feinstein and Reiss 1998; Belmonte and Bourgeron 2006) and tuberous sclerosis (Smalley 1998) have increased evidence of a genetic basis for ASD. Twin and family studies showing high heritability also strongly suggest an important genetic component (Bailey *et al* 1995).

Structural MRI studies have also shown changes suggesting a neuro-biological basis for the disorder, with findings such as enlarged ventricles and differences in some brain areas including the amygdala and the cerebellum (Aylward *et al* 1999; Howard *et al* 2000), as well as reported differences in grey and white matter density (Spencer *et al* 2006). An overall increase in brain size of 2 -10% has also been reported (Piven *et al* 1996). Functional MRI studies have shown differences in brain activation particularly in the fusiform gyrus in relation to face recognition (Schultz *et al* 2000), which may be linked to the social communication deficits such as lack of eye contact often noted in autism. Although there is current rapid development of new technologies and analysis techniques which have contributed to a greater understanding of the exact nature of brain abnormalities and the genetic aetiology in ASD, this remains the subject for much further research.

2.2.7 Diagnosis

As yet there are no confirmed genetic or biological markers for ASD therefore diagnosis must be made on the basis of behavioural observation and developmental history. The diagnostic criteria of DSM IV and ICD 10 have underpinned several

diagnostic tools such as the Autism Diagnostic Interview-Revised (ADI-R; Lord *et al* 1994), the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing *et al* 2002), both with parent interview format and the Autism Diagnostic Observation Schedule (ADOS; Lord *et al* 1989) which involves direct observation of the individual. The Social Communication Questionnaire (SCQ; Berument 1999) was developed by the same team as the ADI-R as a screening questionnaire for use with parents; it is quick to use requires little training and shows good agreement with the ADI-R.

Although ASD is present from birth it is often not obvious in the earliest months of development and concerns are not usually raised until the child is at least 18 months to 2 years old, and often much later than this. In the case of AS, diagnosis may not be given until adulthood, if at all (Tantam 1991). Brogan and Knussen (2003) found an average time delay of 3 years (ranging from no delay to 12.7 years) between first suspicions and final diagnosis of ASD in their sample of Scottish parents, although there was no significant association between time delay and satisfaction with disclosure of diagnosis of autism. Howlin & Asgharian (1999) found that children with autism received a diagnosis on average at age 5, whereas for those with AS average age at diagnosis was 11 years. Parents in this study had been aware of their child's difficulties since an average age of 18 months for autism and 30 months for AS and expressed a degree of frustration at the time delay in receiving a diagnosis.

Bristol (1987) studied a group of families of children with autism or communication impairment before formal evaluation and found that severity of disability was associated with improved marital adjustment. An explanation for this finding was that increased uncertainty and ambiguity in cases of less severe and less obvious disability had a negative effect on family interaction and contributed to poorer outcome in terms of the marital relationship. In the case of AS, parents have reported their child's condition being unrecognised for a substantial period and mothers in particular, report being given complicated and inaccurate information by clinicians during this time and feeling blamed for their child's condition (Gray 2003). For example, mothers have reported being told that their child's behaviour is an

emotional reaction to their own over-anxiety (Tantam 1991). These findings suggest that having a family member with an undiagnosed ASD may be a source of stress to parents and the family. This is discussed more fully in a later section.

2.2.8 Gender ratio

An overall male to female ratio of 3.8:1 has been found with the proportion of females affected being higher amongst groups with more severe learning disability (Ehlers and Gillberg 1993; Fombonne 1999). In the case of AS, male to female ratio has been reported as 8:1 (Ehlers and Gillberg 1993). The reason for the preponderance of males is not clear, although it has been suggested that males may have a lower threshold than females for brain dysfunction autism and that for a female to show features of autism she could be expected to have a more severe disability; this latter theory is supported by the increase in proportion of females affected with more severe ID (Wing 1984). It has also been suggested that differences can be explained by the 'extreme male brain theory' (Baron-Cohen and Wheelwright 2003). Differences in the processes of empathizing and systemising amongst unaffected females, unaffected males and people with AS show a pattern of females scoring higher than males and males higher than people with AS in empathising tasks and the reverse pattern for systemizing tasks, suggesting that ASD is an extreme expression of the male brain.

2.2.9 Co-morbidity

ASD may co-exist with other medical conditions in up to 25% of cases (Gillberg and Coleman 1996). As well as ID, these conditions include epilepsy, thought to affect up to 33% (Gillberg and Steffenburg 1987), and chromosome and genetic disorders such as Down syndrome, fragile X syndrome and tuberous sclerosis (Gillberg 1998). Mental health problems are also reported to be more prevalent in individuals with ASD although published evidence is scarce (Tonge *et al* 1999). Attention deficit hyperactivity disorder (ADHD), obsessional compulsive disorder (OCD), mood disorders, bi-polar illness and psychotic illness have all also been reported in people

with ASD (Ghaziuddin 2005), as well as a higher rate of behaviour disorders than for other diagnostic groups (Blacher and McIntyre 2006).

Although ASD often co-occurs with ID, the relationship between the two has been contested. Although the original description of children with ASD by Kanner (1943) suggested that most affected children would be of normal or superior intelligence, early studies found that ASD did co-exist with all levels of ID (Wing and Gould 1979) and in fact high rates of around 75% of young people with ASD having ID (IQ < 70) have been suggested (Joseph 2002). However such estimates have been criticised for representing clinic populations rather than the whole spectrum of ASD and an English population study found that only a little above a half (55%) of children with ASD (including AS) had IQs below 70 (Baird *et al* 2006).

2.2.10 Treatment and interventions

Drug treatment does not affect the underlying features of ASD but may be of limited usefulness to control behaviour problems, anxiety or depression (Ghaziuddin 2005). The most useful interventions seem to be behavioural programmes such as Applied Behaviour Analysis (ABA) which teaches desired behaviours or skills in a series of highly structured steps based on repeated prompts and reinforcers (Lovaas 1987). These techniques have been shown to have good results but require intensive and sustained therapy sessions and have been criticised for teaching behaviours in a way that does not represent natural interactions and therefore is not easily generalised into the natural environment (Schreibman *et al* 1991). Such programmes are usually home-based and depend on a high level of commitment from therapists and parents and are therefore likely to have implications for all family members (Williams and Wishart 2003). The American structured teaching programme 'Treatment and Education of Autistic and Related Communication Handicapped Children' (TEACCH; (Schloper 1997) aims to teach skills based on the learning profiles of this population with an emphasis on adapting the environment and helping the individual to function in society through, for example, supported employment programmes (Keel *et al* 1997).

Many suggestions for dietary supplements or restrictions and links with digestive disturbances have been put forward. However, claims that expensive treatments may produce spectacular results may add to the stress of families who in the hope of a 'cure' may invest considerable time and money into a treatment programme with very little scientific evidence of its efficacy (Howlin 2002)

2.2.11 Outcomes

Outcomes for people with ASD are very varied (Kanner 1943; Seltzer *et al* 2004)), with adolescence proving a particularly challenging period with issues such as transition from school and from children's into adult services (PHIS 2001). Some adapt well and have successful careers and family life, but these tend to be the more able AS individuals who have insight into their condition and to a degree have learned compensatory skills (Seltzer *et al* 2004). For others, the prospect is a lifetime of dependency with a tendency to develop mental health problems such as anxiety and depression and who suffer life-long from social isolation (Brogan 2000).

Evidence has suggested that parenting a child with ASD is more stressful than parenting a child with another type of learning or physical disability or mental health problem (Wolf *et al* 1989, Fombonne, 2001; Fombonne *et al* 2001; Eisenhower *et al* 2005). Child characteristics such as challenging behaviour have been suggested as an explanation for this finding (Maes 2003). Another suggestion has been the possibility of the presence of a behavioural phenotype related to ASD, increasing vulnerability to anxiety and depression in the mother (Duarte *et al* 2005). In addition possible negative effects on siblings of children with ASD have also been suggested (Marciano and Scheuer 2005).

Outcomes for individuals and families are likely to be affected by many factors such as IQ level, presence of additional medical or psychiatric disorders and the availability of appropriate interventions and support (Howlin 2002; Volkmar 2002; Bromley *et al* 2004). Gillberg and Steffenberg (1987) found that young people with higher IQ (>50) and those who develop communicative speech before the age of six years have considerably better outcomes in adulthood. The group of young people

with lower IQ, without speech and those who had additional chromosomal abnormalities had poorer outcomes with a higher likelihood of deterioration or temporary aggravation of symptoms and the development of epilepsy around puberty. The issues of family stress, quality of life and support needs for young people with ASD and their families are considered in more detail in the following sections.

2.2.12 Summary

There are increasing numbers of people being diagnosed with ASD, although this number is far short of the expected prevalence, suggesting that there may be substantial numbers of people with undiagnosed ASD (PHIS 2001). Families of children and young people with ASD have been shown to suffer higher levels of stress than families of children with other conditions and mothers report stress and dissatisfaction related to a delay in getting a definitive diagnosis of ASD. This suggests that families where a young person has an undiagnosed ASD may suffer negative effects as a consequence of the lack of explanation for the young person's difficulties and the lack of appropriate services.

This leads to the question: Are there differences in perceptions of stress, family quality of life and usefulness of service provision amongst families of young people with (a) diagnosed ASD and ASN, (b) undiagnosed ASD and ASN and (c) without ASD but with ASN?

2.3 Stress and Coping

This section will review the literature on stress and coping in mothers of children and young people with ID and ASD. Research reviewed focuses on the causes of stress as well as on the processes involved in coping, and on the relationship of potential stressors to each other and to outcomes. Reported differences between coping in mothers and fathers will be discussed, as well as the effect on families. Different approaches to family research will be explored. The overall consensus that there is

increased stress for this group will be highlighted and linked with issues related to quality of life, provision of services and support, and diagnosis of ASD.

2.3.1 Increased stress in mothers and families

Mothers of young people with ID have been widely reported to experience higher levels of stress than mothers of typically-developing young people (Dyson 1993; Browne and Bramston 1998; Stores *et al* 1998; Nachshen and Minnes 2005). Having a child with ASD has been associated with even higher levels of maternal stress than having a child with another developmental disability or psychiatric condition (Wolf *et al* 1989; Fombonne *et al* 2001; Eisenhower *et al* 2005). Factors suggested as contributors to increased stress in caregivers and families of children or young people with ID include: the severity of the child's disability; presence of co-existing psychiatric or behavioural problems (Hoare *et al* 1998; Lecavalier *et al* 2006) resources available to the family; marital quality; socio-economic factors and the family's coping style (McIntyre 2002; Emerson 2003a; Maes 2003; Nachshen 2003; Saloviita 2003; Kersh *et al* 2006).

Most research has considered stress from the point of view of the mother (Failla and Jones 1991; Stores *et al* 1998; Emerson 2003a; Bromley *et al* 2004; Duarte *et al* 2005; Hassall *et al* 2005; Blacher and McIntyre 2006). However, several studies have included the perspectives of mothers and fathers, considering either as care-giver or both as joint care-givers (Dyson 1993; Grant *et al* 1998; Baxter *et al* 2000; Lecavalier *et al* 2006) or comparing their perspectives or responses (Wolf *et al* 1989; Heaman 1995; Olsen *et al* 1999; Saloviita 2003; Hastings *et al* 2005). In comparative studies, gender differences have been reported in terms of perception of stress and in coping strategies identified and used by mothers and fathers (Heaman 1995; Grant and Whittell 2000; Gray 2003; Saloviita 2003). In most cases mothers have had poorer outcomes in terms of psychological distress (Wolf *et al* 1989; Thoits 1995). It has further been suggested that, as well as suffering higher levels of stress, there are also qualitative differences in the experiences of mothers. Hastings *et al.*(2005) reported that mothers experienced more stress and also more depression

than fathers and that mothers' stress was related to partner mental health; fathers' stress was not related to child behaviour or to mothers' mental health.

Grant & Whittell (2000) found that fathers seemed to defer to mothers as primary care-givers with mothers expressing more self-belief and confidence in their caring role than fathers and also using more coping strategies. The use of a greater number of coping strategies by mothers than fathers has also been found in other studies (Thoits 1995; Gray 2003). These findings are compatible with the traditional situation where the mother usually fulfils the role of primary care-giver, taking more responsibility for managing the family and making decisions regarding care of the child. In contrast, fathers are more likely to spend more time outside the home and thus be affected by a broader range of influences. This would explain mothers' increased use of coping strategies and increased levels of stress and also the association of maternal stress with child behaviour and partner mental health. Mothers are also more likely to make greater sacrifices in terms of their career and social life than fathers (Shearn and Todd 2000; Gray 2003; Todd and Jones 2005). The mediating effects of maternal characteristics such as coping style and the perceived level of support, as opposed to the observable number of supports available, have also been recognised (Milgram and Atzil 1988; Wolf *et al* 1989; Olsen *et al* 1999; Greenberg *et al* 2004; Hassall *et al* 2005; Hastings *et al* 2005).

2.3.2 Stress, ASD and challenging behaviour

Child characteristics such as challenging behaviour or communication difficulties, both associated with ASD, have been reported as major additional contributors to maternal stress (Maes 2003). Blacher & McIntyre (2006) found that although mothers of young adults with ASD suffered higher levels of stress and depression than mothers of young adults with Down syndrome, cerebral palsy or non-specific ID, group differences in stress and depression were accounted for by measures of behaviour problems. Hastings *et al* (2005) likewise found that maternal stress in mothers of children with autism was related to child behaviour problems but not to severity of ASD symptoms. Baker *et al* (2003) also found that once behavioural difficulties had been accounted for, level of disability of pre-school children with ID did not contribute further to parental stress while Kersh *et al* (2006) found that child

behaviour in 10 year-old children with developmental disabilities predicted parental well-being. These findings highlight the importance of considering the effect of behavioural difficulties on stress in families of young people with ID and ASD.

In addition to associations with challenging behaviour, other possible reasons for increased stress in families of young people with ASD are suggested by the literature. The difficulties in diagnosing and categorising ASD may contribute to higher levels of stress in affected families (Bristol 1987; Brogan and Knussen 2003). The stage of 'not knowing' or waiting for diagnosis has been reported retrospectively as a time of increased stress (Tantam 1991; Gray 2003). Grant & Whittell (2000) found that families with a member with ID particularly benefited from contacts with other parents and valued and were empowered by the opportunity to discuss experiences and place their own family experience in the context of a shared experience of a similar group.

The socializing deficits in ASD may make the disorder more apparent during adolescence, a time when relationships with peers take on particular significance (Schneider *et al* 2006). This may lead to added stresses for the adolescent with ASD and for the family, particularly if there is no diagnosis and if the young person has AS or high functioning autism and is therefore likely to be in a mainstream educational placement with higher social expectations (Gray 2003). However, there are no reports in the literature comparing the impact of having a family member with an undiagnosed ASD with the situation where ASD has been diagnosed nor any studies which have explored the implications for individuals and families of this situation.

In view of the acknowledged genetic component in ASD, it has also been suggested that in some families parents themselves may be affected by the broader autistic phenotype and that because of this some mothers may be more vulnerable to stress and anxiety-related difficulties (Duarte *et al* 2005).

2.3.3 Research approaches to stress and coping

Different models have been proposed to explain the mechanisms of stress and coping, representing the various underpinning theories that have evolved. There is a general consensus about increased levels of stress in families of a child or young person with ID or ASD but a range of approaches have been put forward to explain the cause, effect and implications for individual family members as well as for the family as a whole. This section will consider stress and coping from the perspective of the three research approaches that have evolved over the last 50 years: burden of care; stress and coping; and most recently positive perceptions. These three approaches will be described and the implications of their contrasting explanations and understanding of stress in families will be explored.

Burden of care

Burden of care approaches, now widely rejected, viewed the family member with a disability as the ‘problem’ and as the cause of stress and looked for ways to alleviate that perceived burden. This approach has been described as the ‘deficit model’ (Grant and Ramcharan 2001). For example, the work of Olshansky (1962) on chronic sorrow depicted parents of children with ID as experiencing on-going and recurring sadness and grief and a sense of loss for the ‘perfect’ child they were expecting. This approach tends to see families as disordered in their functioning or as passive recipients of services representing a problem to be solved by the service provider.

Early stress measurements from this approach consisted of the objective enumeration of life events for example: chronic illness, bereavement or the presence of a child with a disability (Dohrenwend *et al* 1978). The difficulty with this type of measure has been a tendency to assess the presence of circumstances which an objective observer considers to be likely to induce stress without considering the differences in individual responses to circumstances. A commonly-used measure of stress the Questionnaire on Resources and Stress (QRS; (Holroyd 1988)) was criticised in this respect by Glidden (1993) who pointed out that measuring the presence of circumstances occurring in families with a disabled member could not predict

families' responses to these potential stressors. For example true or false responses to statements such as '_____needs a wheelchair', '_____knows his/her own address' or '_____has some unusual habits which draw attention' could all contribute to higher stress scores on this scale without considering the caregiver's perception of stress in response to each of these circumstances. Grant et al (1998) also questioned the validity of objective measures of circumstances which do not take account of individual appraisal after finding that mothers of people with ID rated many caring activities as neither stressful nor rewarding, in other words as having no effect on their perceived levels of stress. This increased recognition of the importance of the subjective experience of stressful situations led to a new approach to research.

Stress and Coping

The stress and coping model looks at the interaction between families' coping methods, available supports and stressful events (Failla and Jones 1991) and recognises that perceived stress is a stronger predictor of negative outcomes than the occurrence of adverse life events (Hassall *et al* 2005). The focus is on coping strategies and carer characteristics such as family functioning, hardiness, care-giver satisfaction and resilience (Failla and Jones 1991; Glidden 1993; Grant *et al* 1998; Grant and Whittell 2000). Failla and Corson Jones (1991) found that families of children with developmental disabilities who cope successfully with on-going stress demonstrate the quality of 'hardiness' which they characterised by four components: 'control, challenge, commitment and confidence'. Such families feel that they have control over their lives, they see meaning in their experience of life events, they take an active part in addressing issues and they see adversity as a challenge they must face, as opposed to a burden they must bear. This research also found that family hardiness, level of family stress, social support and maternal age (older mothers suffering more adverse effects) were associated with maternal satisfaction with family functioning.

Grant & Whittell (2000) found that parents of people with ID successfully employed a wide range of coping strategies. They divided strategies identified through

interviews into those relating to: a) managing events, b) managing meanings and c) alleviating stress with parents using both behavioural and cognitive coping strategies in each grouping. The most helpful strategy in the managing events group was 'relying on your own experience and expertise', in the managing meanings group it was 'realising there is always someone worse off than yourself' and in the alleviating stress group it was 'keeping a little free time for yourself'.

Coping strategies have been suggested to change over time, although most research has been cross-sectional rather than longitudinal (Gray 2006). Increased stress has been reported at times of transition in the family life cycle, for example during adolescence (Wikler 1986). Schneider et al (2006) suggested that family coping during adolescence could be conceptualised as the family's attempt to maintain a meaningful family routine by accommodating internal and external factors.

A frequently used model of stress coping is the Double ABCX model (McCubbin and Patterson 1983), based on the earlier ABCX model (Hill 1971). The ABCX model considers 'A' as the stressor event, 'B' as the family's available coping resources, 'C' as the meaning of the situation for the family and 'X' as the outcome. The double ABCX model adds the concept of adaptation to crises over time. Research using this model has provided further evidence for the mediating role of coping strategies and cognitive appraisal of the situation in successful adaptation to the pile up of stressors (Bristol 1987; Saloviita 2003). Within this model, crisis is understood as occurring when the family's ability to cope with external events and circumstances cannot meet the current demands.

However, research within the stress and coping approach has been criticised for focusing only on negative outcomes and not asking questions about positive perceptions (Baxter *et al* 2000; Hastings and Taunt 2002) and this has led to the emergence of a positive perceptions approach.

Positive Perceptions

The 'positive perceptions' approach has emerged over the last decade (Hastings and Taunt 2002; Dykens 2005) and focuses on the benefits an individual with a disability may bring to their family and the successful coping of some families. A review by Blacher & Hatton (2001) concludes that while families continue to struggle with some aspects of parenting a child with ID there are many reports of positive outcomes. Studies exploring the experiences of families who adopt children with ID report satisfying experiences and benefits from the presence of the child within their family (Glidden 1993). Research which has included measures of positive perceptions have found them to be independent of stress (Hastings and Taunt 2002). Hastings et al (2005) found that mothers of children with autism reported more positive perceptions than fathers despite higher stress levels although positive perceptions were not predicted by child or partner variables such as child behaviour or partner mental health.

The positive perceptions approach encompasses the aim of identifying coping strategies used by successfully coping parents or families. For example, a group of successfully coping parents were investigated by Wai- Ping Li Tsang et al (2001) and described as being: 'self-confident, positive, pro-active, sociable and outgoing, accepting of their child, involved in advocacy, educated, with supportive spouse, valuing the present, with stable family and being financially secure'. Research into the mechanisms of successful and positive coping is aimed at helping to direct appropriate support to families struggling to meet the demands of their caring role (Hastings and Taunt 2002).

Another change in the approach of stress research has been a move from the individualised view of the child as source of stress and the individual family members' well-being as outcomes, to a conception of the family as a system, with each member having an effect on each other, and an emphasis on family functioning as a whole. This approach is informed by systems theory (Von Bertalanffy 1968), described as a science of 'wholeness' which emphasises the inter-dependence of component parts within a system, and has greatly influenced theories of family

therapy and family nursing (Whyte 1997). The validity of this approach was demonstrated by Baxter et al (2000) when they found that stress attributed by parents to their child with ID was predicted by the stress attributed to the youngest sibling without an ID, suggesting that stress relating to the child with ID could in fact be a measure of general family stress. The importance of considering the family perspective was also demonstrated by Giallo and Gavidia-Payne (2006) who found that sibling adjustment was predicted by levels of family resilience rather than by the sibling's individual experiences of stress. These findings highlight the importance and inter-dependence of well-being for all family members as well as suggesting the potential benefits of interventions aimed at building on family strengths.

Positive perceptions along with systems approaches are becoming much more influential in family research as a more holistic view of disability becomes accepted. It has also been suggested that focusing on positive aspects such as the coping strengths of mothers and families is empowering and more likely to engender a culture of support and respect for the parents as the experts on their child. Strengthening parents' positive perceptions may also increase positive coping strategies and in itself reduce stress (Krauss 1993).

Empowerment is often cited as an important goal in support provision (Scottish Executive 2000a), but there may also be negative impacts if parents see the responsibility of negotiating with an unwieldy service provision system for their child's rights as burdensome. For example, Nachshen and Jamieson (2000) examined the links between advocacy, stress and quality of life in a qualitative study of Canadian parents of children with developmental disabilities. They found that although empowerment was a driving theme in service provision, advocacy was associated with negative as well as positive outcomes in terms of stress and family quality of life. Advocacy which was perceived to be effective in achieving goals was felt to be positive, whereas advocacy which involved sacrifices in terms of time and personal commitment and did not result in desired outcomes was found to be a contributor to increased stress for parents.

2.3.4 Summary

Increased stress for mothers of young people with ID and with ASD has been recognised. Possible reasons include: child behaviour, additional presence of ASD, mothers' perceptions of the situation and their use of coping strategies. Mothers have been identified as experiencing more negative outcomes in terms of stress while also showing more positive perceptions in terms of coping. Both of these findings are likely to be due to mothers' central role as primary care giver. Research has moved from a 'burden of care' approach to a stress and coping model which considers the stressors and mediating factors which affect the outcome of family adaptation and is now moving towards a positive perceptions approach which consider the positive aspects as well as difficulties in families' experiences. The focus is on developing an understanding of successful coping as a model to inform the development of appropriate support and interventions for families who experience difficulties.

In the study to be reported in this thesis, the issue of family stress and coping will be approached with reference to the stress and coping model, with the stressors of child behaviour and the presence and severity of ASD being assessed. The mediating effects of coping strategies and meanings attributed to the situation will be assessed using the Family Stress and Coping Interview (FSCI; (Nachshen 2003)). This questionnaire was developed to address the issues of stress, coping and positive perceptions by asking parents directly about the level of stress experienced in relation to a set of issues derived from focus group research with a parents of children with developmental disabilities. A set of qualitative questions is included, allowing the parent to explain their responses. The addition of questions asking about successes and positive outcomes as well as challenges and difficulties, along with the inclusion of groups of parents in focus groups to identify relevant issues for the scale being developed, represents an attempt to include the perspective of service users by recognising their expertise and valuing their opinions (Nachshen 2003).

The outcomes of individual and family quality of life will be assessed using the Family Quality of Life Survey (FQoL; (Park 2003)). Positive perceptions will also

be assessed using the qualitative section of the FSCI which gives the respondent the opportunity to talk about successes and achievements as well as identifying unmet needs. This approach recognises that stresses and unmet needs can co-exist with positive perceptions and rewards from the care-giving role.

2.4 Quality of Life

This section will describe the development of Family Quality of Life (QoL) as a concept and review research on individual QoL and QoL in relation to ID. Reported debates on the definition of QoL and identification of relevant domains will be discussed. The issues of subjective and objective assessment, the validity of satisfaction as an indicator of QoL and cross cultural aspects will also be explored. Research investigating experiences of families with young people with ID and other disabilities will be reviewed. Links with provision of services and support, maternal stress and issues relating to ASD will be highlighted.

2.4.1 Definition of QoL

There is general agreement that QoL encompasses a number of broad domains that contribute to the subjective degree of well-being experienced within a cultural and social context. For example, Schalock et al (1989) emphasised the subjective and objective components of QoL as reflecting well-being in a cultural context and further suggested that QoL should be the basis for evaluation of interventions and supports. They described QoL as:

‘the degree of independence, productivity, and community integration that a person experiences as determined by subjective reports or objective evaluations’. (Schalock *et al* 1989)

The World Health Organisation more recently defined QoL as:

‘...individuals’ perceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards and concerns....’ (WHOQOL Group 1998)

Although QoL has been defined in different ways, there is general agreement about the main concepts. The QoL Special Interest Research Group (SIRGQL) of the International Association for the Scientific Study of Intellectual Disability (IASSID) conceptualised QoL according to four principles, defining QoL as:

- I. being multi-dimensional and influenced by personal and environmental factors and their interactions
- II. having the same components for all people
- III. having both subjective and objective components, which should be measured by a range of research methods as appropriate
- IV. being enhanced by self-determination, resources, purpose in life and a sense of belonging (Cummins 2005)

There is also agreement that QoL should be measured in terms of domains, each domain having a related set of indicator variables (Verdugo *et al* 2005). Although there are variations in the domains chosen, the over-riding concept is that the set of domains should together encompass all aspects of the construct of QoL. Domains identified as relevant include: physical/ material, psychological/emotional, level of independence/ability to be productive or creative, social relationships, environment and spirituality/personal beliefs (WHOQOL Group 1995; Felce 1997; Schalock 2004). Indicator variables are identified in relation to each specific domain and are related to behaviours, conditions or reported perceptions that represent relevant quality outcomes. Choosing appropriate indicator variables whether measuring subjective or objective experiences involves judgements about the importance of respective issues and their potential use in improving outcomes (Verdugo *et al* 2005).

Cummins (1995) proposed a multi-dimensional model with seven domains, his scale measured satisfaction weighted by reported importance for each domain. This approach recognises the mediating effect of the importance attached to aspects of QoL that may differ between individuals. Park *et al* (2003) used a similar model in their Family QoL scale which rated each item across five domains for satisfaction and importance. The inclusion of a measure of importance ensures that families' priorities are represented in any evaluation of QoL.

The SIRGQL group has also argued against the development and use of QoL measures for specific groups or conditions. They suggest that such measures tend to treat the absence of pathology relating to a specific condition as an indicator of higher QoL and similarly they tend to include condition-specific indicator variables that cannot be measured against population norms as they have no relevance (Cummins 2005). QoL measures developed for specific groups such as people with particular conditions or disabilities or measures such as health-related QoL (Allik *et al* 2006) may be affected by these limitations in that they do not take into account the whole life experience of the individual or family and that increasing severity of a health problem may not necessarily equate with a decrease in QoL. SIRGQL argue that QoL has universal dimensions which apply to all people and that generic measures of QoL should be used (Verdugo *et al* 2005).

Cultural context

The importance of cultural context in explorations of QoL has been recognised (Skevington *et al* 2004; Aznar and Castanon 2005; Schalock 2005) after much debate about whether there could be a cross-cultural consensus about the conceptualisation of QoL. The World Health Organisation (WHO) identified 4 domains: physical, psychological, social and environmental for their WHOQOL-BREF, a short form of the WHOQOL 100 (WHOQOL Group 1995; WHOQOL Group 1998) which also proved to be cross-culturally valid (Skevington *et al* 2004) although slightly different versions were developed for use in different cultures, for example, the Taiwan version (Wang *et al* 2006b)

2.4.2 QoL as a subjective v. objective measure

There has also been debate about whether QoL should be a subjective or an objective measure or a combination of both. If both, then the question arises of how much weight should be added to the subjective and objective elements. In particular, the issue of subjective well-being (SWB) has been identified as a QoL indicator which might have a global influence whether positive or negative on satisfaction in other domains (Cummins 2005). It has been suggested that an adaptive psychological mechanism will tend to maintain satisfaction at a certain level in spite of changes in

circumstances (Cummins 1995), with people generally tending to view themselves as having a better quality of life than others around them or, in difficult circumstances, as coping better than others or better than could be expected. After reviewing 15 studies of life satisfaction in adults, Cummins suggested that people tend to report satisfaction within a narrow range, with most people rating their SWB above the mid point (50%) on average maintaining this level at around 75%. This means that most people rated their satisfaction with life as being significantly above average. Cummins suggests that SWB may remain stable over time due to this homeostatic mechanism which would make SWB, often characterised as 'satisfaction', a less sensitive indicator of QoL. Although it is claimed that measures of satisfaction may only be sensitive to major changes, are related to disposition and are subject to psychological regulatory mechanisms, it is possible that they may be useful where satisfaction is particularly high or low. Lowered SWB is likely to be a sign of sudden and/or serious negative change in circumstances or the onset of clinical depression (Cummins 1995). SWB is therefore considered to be the least sensitive variable but also the most important because of the significance of changes when they do occur.

2.4.3 QoL for people with ID

The concept of QoL assessment has gained increasing recognition since the early 1980s when principles of 'normalisation' and 'social role valorisation' (Wolfensberger 1983) reflected the growing ethos of the time towards recognising and promoting the rights of people with ID to equality and inclusion within society. QoL has served as an over-arching theme across various disciplines and professional boundaries in the development of a framework for service provision for people with ID (Schalock 2004). QoL is suggested as an appropriate a goal of service delivery as it reflects the shift in emphasis from outcome in terms of individual aspects such as physical health or wealth towards an understanding of the interdependence of social and inter-personal well-being in the assessment of positive outcomes (Schalock 2004). It has been suggested that enhanced QoL for individuals and families may be the only acceptable outcome of services and policies (Osher 1998).

The lack of sensitivity of satisfaction as a QoL indicator may be particularly relevant for people with ID. Felce (1997) suggested that use of subjective indicators of QoL for people with IDs may fail to identify areas of unmet need for this group and that objective standards for the adequacy of support provision should also be considered in any evaluation. Responses on QoL measures may also be affected by a degree of social desirability in the answers given, reflecting how the responder wishes to be perceived by others (Hensel 2001). When applied to people with ID there may be a confounding factor of acquiescence (Heal and Sigleman 1996), making satisfaction a difficult concept to assess in measurement for QoL for this group.

Another suggested limitation may lie in the definition of QoL itself as this may involve one group imposing their values and deciding what constitutes a life of quality for others. There is no doubt that in general professionals are setting the agenda in QoL research rather than people with ID (Northway and Jenkins 2003). For example, in Schalock's definition of QoL quoted at the beginning of this section the inclusion of 'independence' and 'productivity' as indicators of QoL has been disputed in relation to people with ID, as relating to a value judgement of others which may not reflect personal experience and perceptions of QoL (Northway and Jenkins 2003). It has also been suggested that measuring QoL may show people with ID having a lower QoL and that this may result in a value judgement inferring that that because their QoL is lower, the value of their life is also lower (Brown 1999).

QoL is an important consideration in the evaluation of services for people with ID because of greater social awareness about issues of quality and growing concern about the quality of life for individuals in community placements (Northway and Jenkins 2003). There is also dissatisfaction with current methods of monitoring and evaluating service outcomes and quality of service and a wider acceptance of user viewpoints in determining satisfaction (Whoriskey 2003). User viewpoints are now increasingly recognised as including the views of carers and family members of people with ID. This approach values the individual's subjective perception of their QoL and recognises the role of family in that perspective. This makes the development of measures of family QoL a natural progression.

2.4.4 Family QoL

The definition of 'family' has changed from a rigid traditional view of two married parents and their biological children living as a family unit to reflect the varied life styles of today's society. A more inclusive definition of family does not depend on biological or legal ties, for example:

‘A family includes the people who think of themselves as part of the family, whether related by blood or marriage or not, and who support and care for each other on a regular basis’
(Poston 2003)

While there is international consensus on the conceptualisation of individual QoL, the issue of family quality of life is at an earlier stage in development. There has been increasing awareness in the UK and internationally of the needs of families of people with ID (Scottish Executive 2000a; Turnbull *et al* 2004) and a move towards a more systems-orientated approach to service provision (Verdugo *et al* 2005). The systems perspective is influenced by systems theory (Von Bertalanffy 1968) and sees the individual in terms of the environments they inhabit. The family system can be understood in terms of stability, change, circularity and boundaries (Whyte 1997). Stability refers to the family's attempt to maintain equilibrium while coping with change and development, circularity refers to the effect that each member's action has on each other member and boundaries are how the family members define themselves within the family and how the family functions in relation to society.

As the focus of service provision has become more community-based and the power balance between service users and service providers has begun to move towards empowering service users, the importance of the family in supporting people with ID has been increasingly recognised by professionals. This encompasses ideas of respecting and encouraging the contribution of service users and family carers and is compatible with person-centred planning in which the individual is seen in the context of his/her individual circumstances (Renwick *et al* 2000). This change in emphasis is reflected in the increase in the number of published papers relating to

family issues in ID over the past 20 years (Glidden 1993; Hatton *et al* 2003; Hastings *et al* 2006).

Very little research has directly addressed the issue of QoL in families of a child or young person with ID however, with only two main studies reported in the literature. Firstly, the 3 country study from Australia, Canada and Israel (Brown *et al* 2003) and secondly, the family QoL study from the Beach Centre for Disability in the USA (Poston 2003). In the 3 countries study, it was found that overall satisfaction remained fairly high in spite of the difficulties faced by these families, a finding thought to be an example of the homeostatic protective mechanism sustaining the perception of well-being (Brown *et al* 2003). Families in the 3 countries study reported lower satisfaction with formal and informal support. The two studies generated similar domains, 9 in the 3 country study and 10 in the Beach Centre study.

Poston and colleagues developed The Family Quality of Life Survey (FQoLS) from quantitative and qualitative research (Park 2003; Poston 2003). FQoL domains and indicators were derived from focus groups and individual interviews with families and children with and without disabilities and with service providers, a qualitative approach was taken due to the exploratory nature of research into family QoL which had been little reported elsewhere. The domains were tested in a large field test and factor analysis reduced the domains and developed the structure. There were 25 items which grouped into 5 factors: family interaction, parenting, emotional well-being, physical/material well-being and disability-related supports. A self-report questionnaire was developed asking respondents to rate the importance of items for their family to have a good quality of life and then their satisfaction for that item. The survey was used in a Spanish population (Verdugo *et al* 2005) and showed good test-retest reliability and internal consistency as well as cross-cultural validity. An American study likewise showed good support for the validity of the constructs measured (Wang *et al* 2006a)

As with other areas of family research, most ID studies have tended to focus on the mother's perception of family QoL. In line with results of studies on stress, depression and other negative outcomes, studies comparing the experiences of different family members in relation to QoL have shown that in general mothers are more adversely affected (Wolf *et al* 1989; Thoits 1995). Few studies have examined the perspective of family members of young people with mild or borderline ID or other ASN such as ASD. However, one study (Allik *et al* 2006) looked at health-related QoL in mother and fathers of young people with AS or High Functioning Autism (HFA) and found that, in common with other groups, mothers in particular reported lower health-related QoL, although fathers were not affected.

There is also mixed evidence about the effects on siblings. While the majority of siblings adjust well to having a family member with a disability (Skotko and Levine 2006), a small number may be at risk of suffering adverse effects (Sharpe and Rossiter 2002) and this may be the case in particular for siblings of children with ASD (Fisman *et al* 2000; Marciano and Scheuer 2005). However, Giallo and Gavidia-Payne (2006) found that sibling adjustment was predicted by family resilience factors rather than individual experience of stress and coping. This highlights the importance of considering the QoL of the family as a whole as well as from the perspective of individual family members.

2.4.5 Summary

While there is an established research base related to QoL in people with ID, family QoL is a much newer concept. The importance of the effect of all family members on each other and on the functioning of the whole family has been recognised and has been considered in service developments. Family QoL encompasses the notion of empowerment and family-centred service provision and is a useful concept in the assessment of service outcomes. In fact it has been suggested that family QoL should be considered as a major outcome measure for evaluation of service provision.

The recognition of the use of family QoL measures as a guide to quality enhancements and evaluation of service and support provision makes it an appropriate choice for comparing outcomes for families of young people with ASN and ASD with and without diagnosis in the study reported in this thesis.

The additional use of measures of maternal stress, child behaviour, services used and satisfaction will allow a more detailed exploration of family outcomes and family QoL. Blacher and Mink (2004) warn of the danger inherent in research that uses a variety of measures with one respondent, describing this issue as ‘shared method variance’. This refers to the situation where the respondent’s responses may be dependent on each other: for example, the mother may report a high level of behaviour difficulty, a high level of stress and a low quality of life but because of the multiple measures it is not clear which of these variables is having the main effect and how much they are influencing each other. A suggested way to avoid this difficulty is to involve other family members in completing the measures, therefore use of a family measure such as the FQoLS may be useful to capture the viewpoints of other family members. The FQoLS (Park 2003) addresses the issue of differing perspectives of family members, with ratings from individuals within the family which can be aggregated as a ‘family score’ or analysed individually to allow comparisons between different family members. Using qualitative interview methods may also help to validate the questionnaire findings by allowing mothers the opportunity to explain their responses in more detail (Blacher and Mink 2004).

2.5 Conclusion

This chapter has drawn together findings and relevant issues from the literature, pointing out gaps in knowledge and suggesting a rationale for the current study. The literature suggests that young people with ASN, their carers and families have unmet support needs and that although new legislation and policies seem to address these needs, there is little evidence as yet for improved outcomes for this group. Particular challenges facing young people with ASN include difficulties with transition to adult

services including patchy post school provision. Social difficulties with making friends and taking part in community activities have also been highlighted.

Carers and family members of young people with ASN suffer increased stress, with those with ASD reported to suffer even higher stress levels. ASD is as yet poorly defined and categorised and diagnosis seems unreliable. There are therefore likely to be good numbers of young people affected by ASD which has not been diagnosed. Challenging behaviour has also been identified as a major contributor to family and maternal stress and is often a major component of ASD.

Family QoL is a fairly new area of investigation and has been informed by individual QoL research and influenced by a systems theory approach, recognising the interdependence of family members and the significance of their interactions for family well-being. It has been suggested that family QoL is the most appropriate outcome measure for service delivery for families of young people with ID. The Family Quality of Life Survey (FQoLS; Park 2003) is viewed here as a particularly appropriate family QoL measure for this study as it takes account of both satisfaction and importance of issues and allows multiple family members accounts to be included (for full methodology see chapter 3).

Use of both quantitative and qualitative methods were considered appropriate to explore different aspects of mothers' and families' experiences. For example, the family stress and coping interview (FSCI; Nachshen 2003) involves a stress rating questionnaire and also a semi-structured interview allowing respondents the opportunity to expand on their responses (see chapter 3). The questions to be addressed are as follows:

- Do mothers of young people with ASD in this study population have higher stress levels than those with ASN but no ASD?
- For young people with ASD what effect does having a diagnosis have on stress levels and maternal and family QoL?

- Could stress be explained by behavioural difficulties or other factors?
- How does maternal stress relate to family QoL?
- How does QoL compare with mothers and families of typically developing young people?
- Does the level of and satisfaction with service provision have an effect on stress and family QoL?
- What are mothers' perceptions of stressful issues for their family?
- What are mothers' perceptions of unmet needs for their family?

CHAPTER THREE - QUANTITATIVE PHASE OF THE STUDY

This chapter contains descriptions of the aims and design of the quantitative phase of the study including: recruitment procedures, variables studied, measures used and their application to the sample group. The results are reported, the analysis described and interpretation discussed. The chapter concludes with a summary of the main results and their implications and provides a rationale for the complementary work of the qualitative phase of the study to be described in the following chapter.

3.1 Aims

The study aims to explore perceptions of mothers of young people with Additional Support Needs (ASN) in relation to stress, service provision and family and individual quality of life and to determine unmet needs. A particular focus is the effect of having a family member with ASN who has an additional diagnosis of Autistic Spectrum Disorder (ASD) and also the situation where the young person with ASN may have an ASD which has not been recognised or diagnosed. Previous research has suggested that maternal and parental stress is higher in families of young people with learning or developmental disability (Stores *et al* 1998) and that parents of children with ASD suffer more stress than parents of children with other forms of developmental disability or a psychiatric condition (Wolf *et al* 1989; Fombonne *et al* 2001). Child characteristics such as challenging behaviour or communication difficulties have been reported as major stressors while the mediating effects of maternal characteristics such as coping style and the perceived level of support have also been recognised (Hassall *et al* 2005).

This phase of the study explored relationships between and amongst the following variables: family QoL, level of child behavioural difficulties, presence and severity of autistic features as determined by score on an autism screening measure, level of maternal stress, maternal QoL and level of support from services.

In particular, the following questions were addressed:

- Is there a difference in family quality of life (FQoL) between families of young people with ASN and ASD and those with ASN but no ASD? If this is the case then why should this be so?
- What are the implications for families of having a young person with ASD that is not diagnosed? Are FQoL, maternal individual QoL or maternal stress affected when there is no diagnosis, either negatively or positively?
- What are the additional effects on FQoL, maternal QoL and maternal stress of: presence of behaviour difficulties, severity of ASD, child age, gender and level of service provision?
- Are there particular significant differences in FQoL, maternal QoL and maternal stress between families of typically developing young people and families of young people with ASN?

3.2 Sample

The sample was recruited from mothers of young people aged 13-22 who were participants in the larger parent study as described in Chapter one.

The study sample consisted of 93 mothers. The young people were categorised as falling into one of four groups:

1. having ASN, a negative ASD screening score but no diagnosis of ASD (41)
2. having ASN, a positive ASD screening score and a diagnosis of ASD (18)
3. having ASN, a positive ASD screening score but no diagnosis of ASD (17)
4. typically developing, unrelated, age-matched controls (17)

The four groups were selected in order to allow comparisons of those young people with possible ASD with and without a diagnosis and to provide insights into the effect of having a diagnosis of ASD on the family variables being studied. Of the young people with ASN approximately half of those recruited scored below the ASD cut-off on the screening measure (the Social Communication Questionnaire SCQ; Berument 1999) and formed group 1, while the half scoring above the cut-off were divided again into groups 2 and 3 (with and without a diagnosis of ASD). Having a

group with ASN and no positive SCQ score allowed exploration of whether any differential effects of ASD diagnosis are due to ASN rather than ASD while comparisons with the typically developing control group allowed exploration of the effects of having a young person with ASN in the family.

3.2.1 Selection

A large number of potential participants were known to the researcher through previous involvement with the parent study and therefore there was some initial selection of the sample for this study in terms of people who were known to be interested in further participation and were therefore likely to agree to complete the questionnaires. Families invited to take part were selected from all areas of Scotland although for practical reasons there was an over-representation of those from less geographically distant areas, for example from Lothian, Lanarkshire and Greater Glasgow. The sample was therefore not necessarily representative and it cannot be known whether the issues identified and concerns expressed were different from those of the wider population of families of young people with ASN. However, the mothers who participated represented a wide range of social backgrounds and varying degrees of involvement with services, the young people themselves had a broad range of ability. The young people in the parent study were recruited from special schools, mainstream schools and from colleges of further education.

3.2.2 Recruitment

Mothers were contacted initially by telephone and asked whether they would be interested in completing the questionnaires. Telephoning before sending the letter of invitation was felt to be useful as it allowed the researcher to explain the study and informally answer any questions, thereby encouraging a positive response. If the mother expressed an interest the questionnaires were sent with a letter of invitation, a consent form and information sheet (see appendix 3). In the case of those attending for their son or daughter's participation in the parent study, mothers were asked if they would be willing to complete the questionnaires while attending. Mothers were asked whether other family members might be willing to complete a questionnaire

and if this was the case the requested number of questionnaires were sent to the family along with a letter of invitation for family members (see appendix 3). It was also explained that a sub-set of mothers would later be invited to take part in an interview.

3.2.3 Consent and ethics

All the mothers and young people had already given their consent to take part in the parent study. Some had been visited at home several times and had attended for testing on one or more occasion and so were well known to the research team. The requested number of questionnaires were sent to the family, a letter of invitation and a consent form for each family member to sign was included. It was clearly stated that there was no obligation to take part. The study protocol, information and consent sheets were submitted to the UK Multi- centre Research Ethics Committee and given approval.

3.2.4 Background data

During the parent study the researcher was involved in the collection of data from the mothers. This included questionnaires about child behaviour difficulties and presence/severity of autistic features as well as background and demographic information about the young people's age, gender, and type of school attended, along with a brief family and obstetric history and information on any diagnoses received. The family's postal code gave a rough indicator of socio-economic status (SES). This allowed variables such as age, gender, health status and SES to be explored and any confounding effects controlled for if necessary. Demographic data is detailed in the following table (Table 3.1)

TABLE 3.1 Demographics of sample

Variable	n	%
Gender		
Male	60	65
Female	33	35
Young person age		
13-15	44	47
16-18	39	42
19-22	8	9
Mother age		
30-40	10	11
41-50	66	71
51-60	11	12
Area		
Lothian	34	37
Lanarkshire	13	14
Renfrewshire	11	12
Aberdeenshire	8	9
Greater Glasgow	16	17
Fife	4	4
Other (Tayside, Borders, Perth, Inverclyde)	7	8
Recruited from		
Mainstream school	45	48
Special school	37	40
Adult services/courses	11	12
Mother's report of young person's diagnosis		
ASD	18	19
No diagnosis	25	27
Learning difficulty/disability	10	11
Dyslexia/dyspraxia	17	18
ADHD	11	12
Epilepsy	3	3
Genetic abnormality	4	4
Other(<i>Tourette's OCD, DAMP, behaviour problem, mild CP</i>)*	8	9

* For full list of abbreviations see appendix 1

3.3 Measures

The following two questionnaires, were completed as part of the parent study, with data consequently available for this study.

3.3.1 Child Behaviour Checklist (CBCL)

The Child Behaviour Checklist (CBCL; Achenbach 1991) gives a behavioural profile for young persons between 13-16 (see appendix 2). This measure provides a score of behaviour in relation to population norms, as well as dividing this into scores for eight domains: withdrawal, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behaviour, and aggressive behaviour. There is evidence that the CBCL has good reliability and validity when used in research with children and adolescents with mild ID (Epstein *et al* 1984; Schachter *et al* 1991; Crijnen *et al* 1999; Dekker *et al* 2002) with findings showing that such children and adolescents are significantly more at risk for emotional and behavioural problems than their typically developing peers.

CBCL score was used to control for child behaviour difficulties when assessing the effect of ASD on maternal stress and family QoL. Previous research has identified the increased prevalence of emotional and behavioural difficulties in young people with moderate to borderline ID (Douma *et al* 2006) as well as the possibility that higher stress levels in parents of young people with ASD can be accounted for by increased levels of behaviour problems (Blacher and McIntyre 2006). The relations between behaviour difficulties and ASD were also explored as previous research has suggested that certain items on the CBCL can successfully discriminate between young people with and without ASD (Duarte *et al* 2003). It was hypothesised that amongst young people scoring above the cut off for ASD, those with high CBCL scores would be more likely to have a diagnosis of ASD than those with lower scores because those with behavioural difficulties are more likely to come to the attention of service providers.

3.3.2 Social Communication Questionnaire (SCQ)

In order to identify a group of young people who may have an ASD which has not been recognised or diagnosed the whole sample was screened with the Social Communication Questionnaire (see appendix 2, SCQ; Berument 1999). The SCQ is an ASD screening tool derived from the Autism Diagnostic Interview- Revised (ADI-R; Lord 1994). A 40-item parent questionnaire, the SCQ is quick and easy to use and has been well validated, showing good discriminative validity with respect to the separation of Pervasive Developmental Disorder (PDD) from non-PDD diagnoses at all IQ levels. Scores are given in three ranges: non-PDD, PDD and autism. The cut-off score of 15 between PDD and non-PDD was most effective, with weaker discrimination between PDD and autism with best differentiation at a cut-off score of 22. In this study the cut-off of 15 was used to distinguish those with a positive screening score for ASD from those with a negative score.

Using the SCQ, large numbers of children can be screened and screening scores compared with maternal report of diagnosis. However, as this is a screening and not a diagnostic tool, further investigations would be required before a diagnosis of ASD could be given in any individual case.

In order to confirm the findings of the SCQ screening and to give useful feedback to participants, as part of the parent study a subset of young people have been assessed with the Autism Diagnostic Observation Schedule (ADOS; Lord *et al* 1989), a standardised protocol for the observation of social and communication behaviour related to ASD based on ICD-10 diagnostic guidelines and mothers have been interviewed with the ADI-R. This work, by other members of the parent study research team, is on-going, results are not yet available and it does not form part of this thesis.

In the quantitative phase of the study being reported here, parents completed a set of additional study-specific questionnaires incorporating measures of the following variables:

3.3.3 Family Quality of Life Survey

Family quality of life was measured using the Family Quality of Life Survey (FQoLS; Poston 2003). This is a self report questionnaire which can be completed independently by multiple family members including the young person with ASN (see appendix 2). Respondents are asked to rate, on a Likert scale, the importance of 25 aspects of family life, and are then asked to rate their satisfaction with that aspect for their own family. In the analysis the overall satisfaction score is weighted by the importance given to each of the aspects involved. This questionnaire was chosen for its ability to capture not just the experiences of mothers but also those of other family members on the grounds that mothers may have a particular perspective that is not necessarily shared by other family members (Hastings 2003; Saloviita 2003; Seltzer *et al* 2004). In this study the main respondents were all mothers. However, by collecting additional data from family members, the study also allows comparison of mothers' with fathers' scores or mothers' with composite family scores.

The following adaptations were made to the original format of the FQoLS with permission from the authors. Firstly, the term 'disability' was replaced with 'additional learning needs' to fit with the terminology used in the earlier stages of the study. This recognised that while the study participants had been recruited from schools where the child received additional support for learning, in many cases the family did not have contact with clinical services. It also reflected the finding that often the young person had no diagnosis and was not considered, by self or family, to be 'disabled'.

Secondly, as the final four items on the scale relate specifically to families of a young person with a disability/ASN these were removed from the questionnaires completed by the group 4 families (typically developing controls). Scores for the final 4 items (disability issues) were compared for Groups 1 to 3, but in comparisons of all four groups, total scores without the final four items were used.

Demographic information requested was restricted to the respondents' gender, year of birth and relation to the young person. As background information about the

family, including medical history and type of school attended by the young person, had already been collected in the main study, it was felt that questions about parental level of education and family income may have been perceived by respondents as unnecessarily intrusive and were therefore removed.

3.3.4 Maternal Individual Quality of Life (Whoqol-bref)

The Whoqol-bref (WHOQOL Group 1998) is a brief questionnaire assessing individual quality of life, developed by the World Health Organisation (see appendix 2). It is a shortened version of the WHOQOL-100 assessment (WHOQOL Group 1995). This measure was included as it is a widely-used and well-validated standardised measure which has been tested in many settings and been shown to have good psychometric properties (WHOQOL Group 1998; Skevington *et al* 2004). The Whoqol-bref can generate a total score as well as scores in four domains: physical health; psychological well-being; social relations; environment. The concept of family quality of life is less well established than that of individual quality of life and as such the FQoL is a newer measure. It was therefore felt that it would be useful at a number of levels to have quality of life measures from both the whoqolbref and the FQoLS as this would allow comparison of the mother's perception of her own individual quality of life with her perceptions of family quality of life while also providing some evidence of level of concurrent validity for the FQoLS.

3.3.5 Family Stress (FSCI)

The Family Stress and Coping Interview (FSCI; (Nachshen 2003), is a 23 item self-report questionnaire and was used to measure maternal stress (see appendix 2). The respondent is asked to rate on a scale of 0-3 the stress currently being experienced in relation to each item. The FSCI is designed for use in families of children or young people with developmental disabilities and as such the content was not suitable for use with the families of typically developing young people; it was therefore used only with groups 1-3. As with the FQoLS, this measure was adapted by a change of the term 'developmental disability' to 'additional learning needs', again to make this

questionnaire compatible with the other measures used in the study. The FSCI also has a qualitative section which was used in the qualitative phase of the study and is described later in Chapter 4.

3.3.6 Level of Support

In order to determine the level of support parents and families were receiving from services, a short self-report questionnaire was developed (see appendix 2). Parents were asked how many contacts there had been with a range of support agencies over the previous four weeks and were asked to rate the usefulness of that contact. This allowed exploration of relationships between level of service contact, maternal stress and maternal and family QoL and also the relationship between this variable and ASD (positive SCQ score) with or without diagnosis.

3.4 Hypotheses

Six hypotheses were formulated:

1. Family QoL (FQoLS score)

The FQoL satisfaction weighted by importance score would be lowest in the group with positive SCQ but no ASD diagnosis, higher in those with ASD diagnosis, higher still in those with ASN and no positive SCQ and highest of all in the control group.

2. Child behavioural difficulties (CBCL score)

Those without ASN would have the lowest scores, those with ASN but no ASD would have higher scores, those with ASN, positive ASD screening but with no diagnosis would have higher still and with those with ASN, positive ASD screening and ASD diagnosis would have the highest scores.

3. Presence and severity of ASD (SCQ score)

Those without ASN would have the lowest scores, those with ASN but no ASD would have higher scores than those with ASN and ASD without a diagnosis and those with ASN and ASD with a diagnosis would have the highest scores.

4. Maternal QoL (Whoqolbref score)

Those with a son or daughter without ASN would have the highest scores, those with ASN but no ASD would have lower scores, those with ASD and a diagnosis would have lower scores still and those with high SCQ scores but no ASD diagnosis would have the lowest scores.

5. Maternal stress (FSCI score: groups 1-3 only)

Those with a son or daughter with ASN but no ASD would have the lowest scores, those with ASN and ASD with a diagnosis would have higher scores and those with ASN and ASD without a diagnosis would have the highest scores.

6. Level of service provision (number of services accessed: groups 1-3 only)

Those with a son or daughter with ASN but no ASD would have the least contact with services, with ASN and ASD, greater contact with services; and those with a son or daughter with ASN and ASD with a diagnosis would have the most contact with services.

3.5 Data Analysis

Data were analysed using the Statistical Package for Social Sciences version 14.0 for Windows (SPSS 2005). Descriptive statistics were used to investigate group characteristics and group differences, including possible confounding factors. Correlations amongst the main variables in the study were analysed using Pearson's correlation coefficient, with partial correlations carried out to control for the effect of one variable while examining the relationship between two other variables. One-way ANOVAs with planned contrasts were used to test for the hypothesised pattern of group scoring for each variable.

In the case of non-parametric data the Kruskal-Wallis test was used to investigate group differences and Spearman's correlation coefficient was used to examine correlations (Field 2000).

3.6 Feasibility Study

Before embarking on the full study, the first cases recruited were treated as a feasibility study. This was carried out to test the methods, in particular:

- mothers' and other family members' willingness to take part
- respondents' ability to complete and return the questionnaires
- whether early data would support investigation of the planned main research hypotheses

Eighteen mothers were invited to take part in the feasibility study and 15 of these agreed. Of those who did not take part, one felt that the questionnaire was not appropriate to their family situation as their son was now older and other children had left home; the other two non-responders agreed by telephone to participate but did not return the postal questionnaire despite follow-up phone calls. In one of the cases the questionnaires were eventually returned after the feasibility study analysis had been completed and data was therefore included in the main study.

The feasibility study showed that methods were practical. Only one change was subsequently required to the layout of the Whoqolbref questionnaire, where a question at the top of a second page was frequently missed. The initial respondents reported that the questionnaires were clear and easy to understand. However, in some cases, mothers said that they would have found it easier to complete the questionnaires with the researcher and this was subsequently offered as an option to all participants.

The feasibility sample was comprised of mothers of 15 young people in 4 groups:

1. ASN with no ASD (3M, 2F)
2. ASN, scoring above the ASD cut-off and with ASD diagnosis (3M)
3. ASN, scoring above the ASD cut-off but with no ASD diagnosis (2M)
4. no ASN (typically developing controls: 3M, 2F)

3.6.1 Analysis of feasibility study data

Small numbers made meaningful analysis of group differences difficult therefore relationships amongst the main variables were examined using Pearson's 'r' correlation analysis. Only groups 1-3 were included in this analysis as the control group had not completed all the questionnaires. Family QoL was significantly correlated with maternal individual QOL and significantly negatively correlated with maternal stress and severity of ASD (SCQ score). Maternal stress was significantly correlated with maternal QoL and SCQ. There were no significant relationships between challenging behaviour (CBCL) and any other variable or between SCQ and maternal individual QoL (WQB). Results are displayed in the following table (Table 3.2).

TABLE 3.2 Correlations amongst feasibility study variables (groups 1-3)

	FSCI	CBCL	FQoLS	SCQ
CBCL	r(10) = .518			
FQoLS	r(10) = -.787**	r(10) = -.17		
SCQ	r(10) = .721*	r(10) = .395	r(10) = -.756*	
WQB	r(10) = -.836**	r(10) = -.494	r(10) = .790**	r(10) = -.625

*p<0.05 **p<0.01

As figures 3.1 and 3.2 below show, there were significant negative correlations between maternal stress (FSCI score) and both Family QoL (FQoLS score) $r = -.787$, $p < 0.01$) and maternal QoL (Whoqol-bref score), $r = -.836$, $p < 0.01$

FIG 3.1. Correlation between maternal QoL and maternal stress

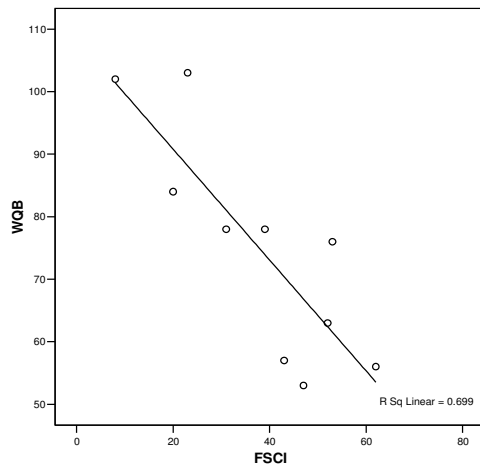
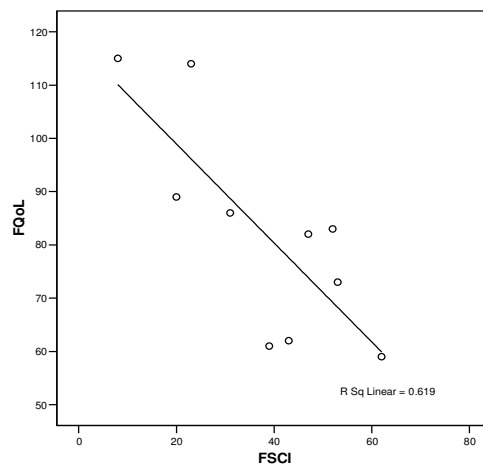


FIG.3.2 Correlation between family QoL and maternal stress



There was also a significant correlation between maternal stress (FSCI score) and ASD screening (SCQ score), $r = .721$, $p < 0.05$ and a significant negative correlation between family QoL (FQoLS score) and ASD screening (SCQ score) $r = -.756$, $p < 0.05$, see figs 3.3 and 3.4 below.

FIG. 3.3 Correlation between maternal stress and SCQ score

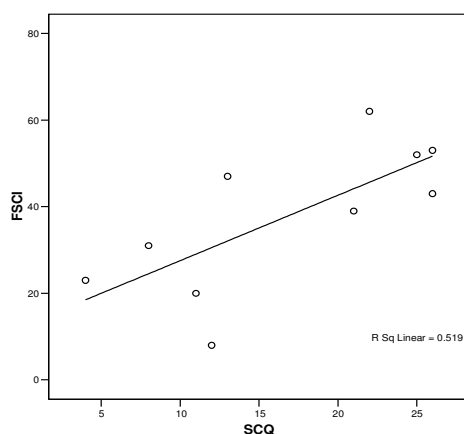
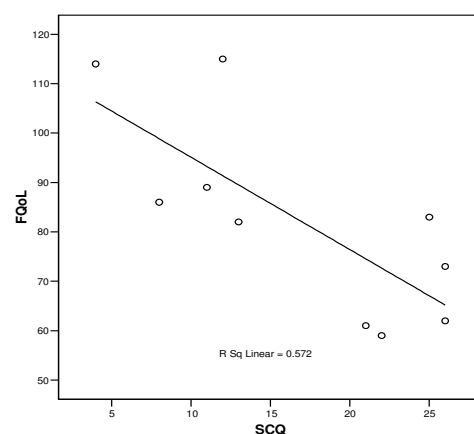


FIG. 3.4 Correlation between family QoL and SCQ score



Results of the correlation analysis suggest that mothers of young people with ASN and ASD are likely to be more adversely affected in terms of individual and family QoL and stress than mothers of young people with ASN but no ASD.

Comparing group means (groups 1-3 with group 4)

Independent t-tests were used to compare group means of group 4 (control group) with group 1-3.

Maternal individual QoL (Whoqol-bref score) was significantly higher in the control group ($p = 0.01$) than the other three groups (Fig. 3.5). Family QoL (FQoLS score) was also significantly higher in the control group ($p = 0.02$) than the other three groups (Fig.3.6).

FIG. 3.5 Maternal QoL for control group compared with groups 1-3

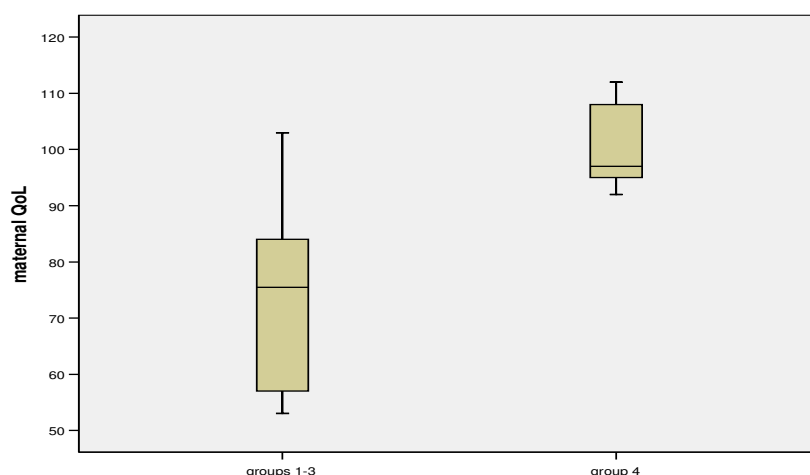
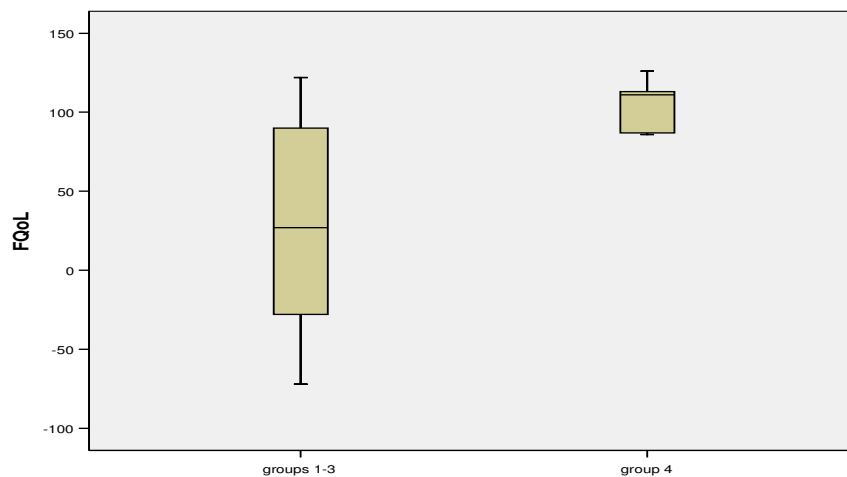


FIG. 3.6 Family QoL for control group compared with groups 1-3



Results of the t-tests suggest that mothers of young people with ASN with or without ASD suffer more adverse effects in terms of individual and family QoL than mothers of typically developing young people.

3.6.2 Results of Feasibility Study

The results suggested that the study methods were appropriate. Statistical analysis partially supported the study hypotheses in that study group profiles showed differences largely in the directions predicted. Following the feasibility study further participants were recruited to the main study as described below.

3.7 Main study

3.7.1 Descriptives

Total numbers recruited in each group were as follows:

1. young people with ASN and negative SCQ score (41)
2. young people with ASN, positive SCQ score and ASD diagnosis (18)
3. young people with ASN, positive SCQ score and no ASD diagnosis (17)
4. typically developing controls (17)

Age

Age range was 13-22 years, with mean age at time of first assessment 15.87 years.

There were no significant differences in mean age amongst the 4 groups

Gender

The control group were fairly evenly divided on gender (47% male), but males were over-represented in all other groups, particularly group 2 (ASD with diagnosis) where there was only one female member of the group.

IQ

IQ scores, measured with the Wechsler Adult Intelligence Scale (WAIS; Wechsler 1999) or the Wechsler Intelligence Scale for Children (WISC; Wechsler 1992) as appropriate, were available for 82 of the 93 young people. There was a significant difference ($p < 0.01$) between the IQ of the control group (mean 111.82) and the other 3 groups. There were no significant differences between mean IQ of groups 1 to 3. The slightly higher mean IQ for group 2 (84.06) than groups 1 or 3 (72.78 and 66.50 respectively) may be due to the inclusion of young people with ASD who were found to have IQs in the normal range or above, although they required and were receiving additional support for educational difficulties.

Table 3.3 provides summary data on these group characteristics.

TABLE 3.3 Group comparisons of age, IQ and gender

Group	N	Mean age(SD)	Mean IQ(SD)	Gender M:F (%M)
1	41	15.60 (1.9)	72.78 (16.86)	23:18 (56)
2	18	16.35 (2.34)	84.06 (22.08)	17:1 (95)
3	17	16.06 (1.40)	66.50 (9.35)	12:5 (71)
4	17	15.85 (1.7)	111.82 (16.88)	8:9 (47)
Total	93	15.87 (1.89)	82.16 (23.33)	60: 33 (65)

Socio-economic status

The socio-economic status (SES) of the four groups was also compared using the ACORN system, a geo-demographic marketing tool (CACI 2003) which uses UK postal codes to rate SES from 1 to 5. The Kruskal- Wallis test showed no significant

differences in mean SES scores amongst the four groups. Chi square was 6.93, sig 0.074, df(3).

3.7.2 Non-responders

In ten cases families did not return postal questionnaires although in each of these cases the mother had agreed by telephone or had taken the questionnaires home after attending with the young person. When a postal questionnaire was not returned, a reminder phone call was made to the mother. In each case the mother again agreed to complete and return the questionnaire but in some cases there was still no reply. In conversation these mothers had expressed willingness to participate but cited being too busy or having forgotten as being the main reasons for their non-response to date and it is presumed that this accounted for those cases in which no reply was ever received. The non-responders were spread across the four groups as shown in Table 3.4 below.

TABLE 3.4 Non-responders by group

Group	Non-responders (N)
1	2
2	2
3	3
4	3
Total	10

3.7.3 School Placement

Young people were recruited into the study through their school or college placement. Mainstream and special schools and colleges of further education were included. While 49% of group 1 attended mainstream school, only 39% of group 2, and 18% of group 3 attended mainstream schools. Percentages attending FE colleges were broadly similar across the 4 groups, although slightly higher for group 3. When those attending college are removed from the comparison, the same pattern remains, with group 1 having 46%, group 2 having 56 % and group 3 having a much higher proportion attending special school at 79%.

Numbers recruited from each setting for each group are shown in Table 3.5 below.

TABLE 3.5 School placement by group

Group (n)	Maintream (%)	Special (%)	FE college(%)
1 (41)	20 (49)	17 (41)	4 (10)
2 (18)	7 (39)	9 (50)	2 (11)
3 (17)	3 (18)	11 (65)	3 (18)
4 (17)	15 (88)	0	2 (12)
Total (93)	45 (48)	37 (40)	11 (12)

3.7.4 ASD Diagnosis by Area

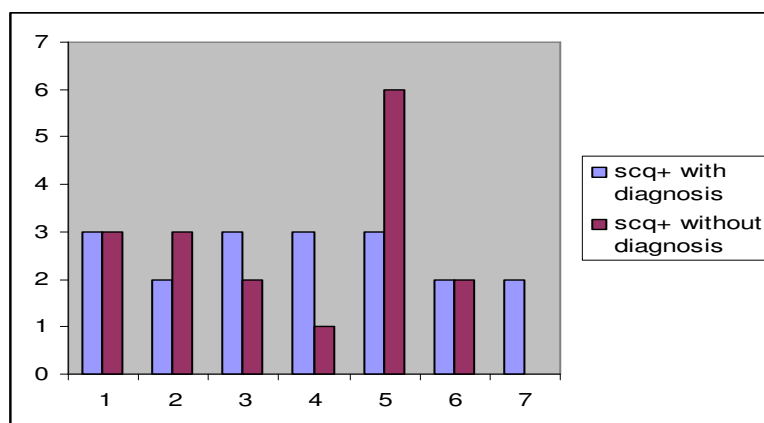
Groups 2 and 3 (scoring above the cut-off for ASD) were recruited from all areas of Scotland and numbers from each area were small. However, groups were compared to look for possible local differences amongst patterns of diagnosis of ASD. Table 3.6 below shows the numbers and percentages of young people scoring above the ASD cut-off with (group 2) and without (group 3) a diagnosis for each of the areas in the study.

TABLE 3.6 ASD status by area

	Area	Group 2 (%) (with diagnosis)	Group 3 (%) (without diagnosis)
1	Lothian	3 (50)	3 (50)
2	Lanarkshire	2 (40)	3 (60)
3	Renfrewshire	3 (60)	2 (40)
4	Aberdeenshire	3 (75)	1 (25)
5	Greater Glasgow	3 (33)	6 (67)
6	Fife	2 (50)	2 (50)
7	Other	2 (100)	0

The groups were fairly evenly divided with only the Greater Glasgow area showing a higher level of high SCQ scores without diagnosis as shown in Fig 3.7 below. Although numbers included are too small to draw any meaningful conclusions, it is possible that local practices are different and that this may be having an effect on the numbers of young people receiving a diagnosis of ASD.

FIG 3.7 ASD status by area: groups 2 and 3



3.7.5 Family members completing FQoLS questionnaire

A range of family members were invited to complete the FQoLS questionnaire (see table 3.7). Although the main respondents were 93 mothers (mean age 45.24), 26 fathers (mean age 47.81) also completed the questionnaire. In addition 15 siblings, 8 brothers (mean age 18.63) and 7 sisters (mean age 17.43) and 22 of the young people on whom the study centred, 11 male and 11 female (mean age 16.31) also completed the questionnaire.

TABLE 3.7 Family members completing FQoLS, Age, Gender

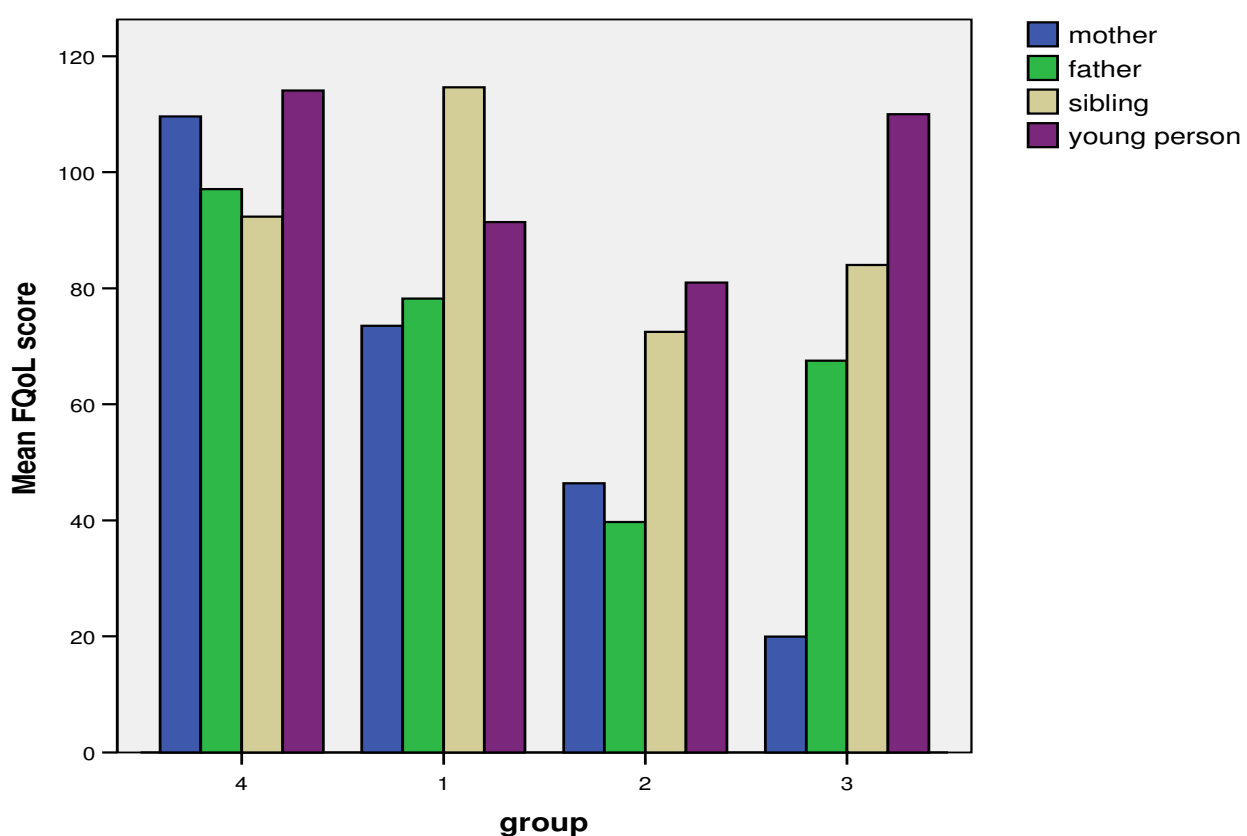
	N	Mean age (SD)	Range	Gender M:F (%M)
Mothers	86	45.24 (4.45)	36, 59	
Fathers	26	47.81 (4.44)	38, 57	
Siblings	14	18.07 (3.86)	13, 27	8:7 (54)
Young people	22	16.31 (2.42)	13, 21	11:11 (50)

The smaller numbers of additional family members completing the FQoLS questionnaire, particularly in group 3, made it difficult to make meaningful comparisons across the four groups, see Table 3.8.

TABLE 3.8 Additional family members completing FQoLS by group

Group	1	2	3	4
Fathers	8	7	2	9
Siblings	3	4	1	6
Young people	5	6	1	10
Total	16	17	4	25

Scores for fathers and young people across the groups showed a similar pattern to scores for mothers, except for group 3 where the groups were the smallest, containing only 1 or 2 members. Scores for siblings did not seem to follow this pattern suggesting that siblings do not have the same perception of family difficulties as their parents or sibling with ASN. Group differences are shown in Fig 3.8 below.

FIG 3.8 FQoLS scores for family members by group

Correlations between family members' scores (see table 3.9 below) showed that mothers' and fathers' scores were significantly correlated ($p < 0.01$). Mothers' and young persons' scores were also significantly correlated ($p < 0.05$). Sibling scores showed no significant correlation with the scores of any other family member.

Although respondents were instructed to complete the questionnaires independently, it is possible that mothers and fathers may have collaborated while completing the questionnaires. It is also possible that the correlation between the young peoples and the mothers scores could be due to the young person receiving help from their mothers in completing the questionnaire and this would be less likely to be the case for siblings.

TABLE 3.9 Family members' FQoLS score correlations

	Mother	Father	Sibling	Young person
Father	$r(26) = .664^{**}$			
Sibling	$r(14) = .289$	$r(8) = .080$		
Young person	$r(22) = .44^*$	$r(15) = .317$	$r(11) = .327$	
Family	$r(35) = .909^{**}$	$r(26) = .829^{**}$	$r(14) = .649^*$	$r(22) = .711^{**}$

* $p < 0.05$, ** $p < 0.01$

3.8 One-way ANOVAs with planned contrasts

The variables below were investigated for differences amongst the four groups using one-way ANOVAs. Data were tested for hypothesised patterns of scoring across the 4 groups for each measure by examining contrasts for linear trend. Normal probability plots were examined for each variable and non-parametric tests were used for exploring data on service contacts and usefulness as distribution was found to be not normal.

Variables

1. FQoLS (mothers' satisfaction weighted by importance)
2. CBCL (with 8 domains)
3. SCQ
4. WQB (with 4 domains)
5. FSCI
6. Service contact and usefulness

3.8.1 FQoLS

(mothers' mean score of satisfaction weighted by importance)

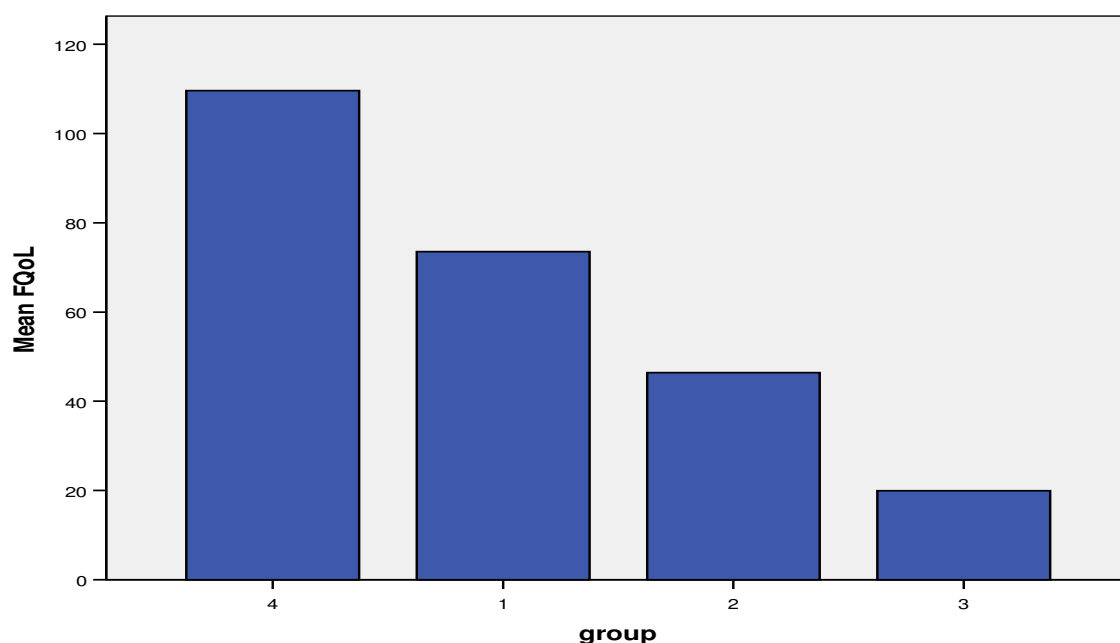
There was a significant effect of group on FQoL ($F(3,83) = 9.88$, $p < 0.01$). Mean scores are shown in Table 3.10 below.

TABLE 3.10 Mean FQoL scores by group

Group	Mean FQoLS	Std Dev	N
1	75.11	42.08	37
2	46.39	67.99	18
3	19.94	60.193	17
4	109.6	23.42	15

The hypothesised pattern of group scores was confirmed using planned contrasts for linear trend ($p < 0.01$), indicating that family QoL decreased proportionately with the control group having the highest QoL followed by group 1 then group 2 then group 3, as shown in Fig 3.9 below.

FIG 3.9 FQoLS by group



Planned contrasts showed that family QoL was significantly higher for the control group than the other three groups ($t(83) = -4.38, p < 0.01$), family QoL was significantly higher for group 1 (non-ASD ASN) than for groups 2 and 3 (ASD with and without diagnosis) ($t(83) = -3.44, p < 0.01$) and there was no significant difference between groups 2 and 3 ($t(83) = -1.56, p = 0.12$)

The mothers of young people in groups 1-3, completed an additional section of the FQoL relating to disability issues. There were 4 issues which were rated for importance and satisfaction in the same way as the remainder of the questionnaire. Results showed that group 1 scored significantly higher than groups 2 and 3 ($p < 0.01$) although groups 2 and 3 were not significantly different from each other, see table 3.11 below.

TABLE 3.11 Mean disability issues score by group

Group	Mean FQoLS(dis)	Std Dev	N
1	14.19	11.20	36
2	1.39	13.04	18
3	3.00	11.65	17

3.8.2 CBCL

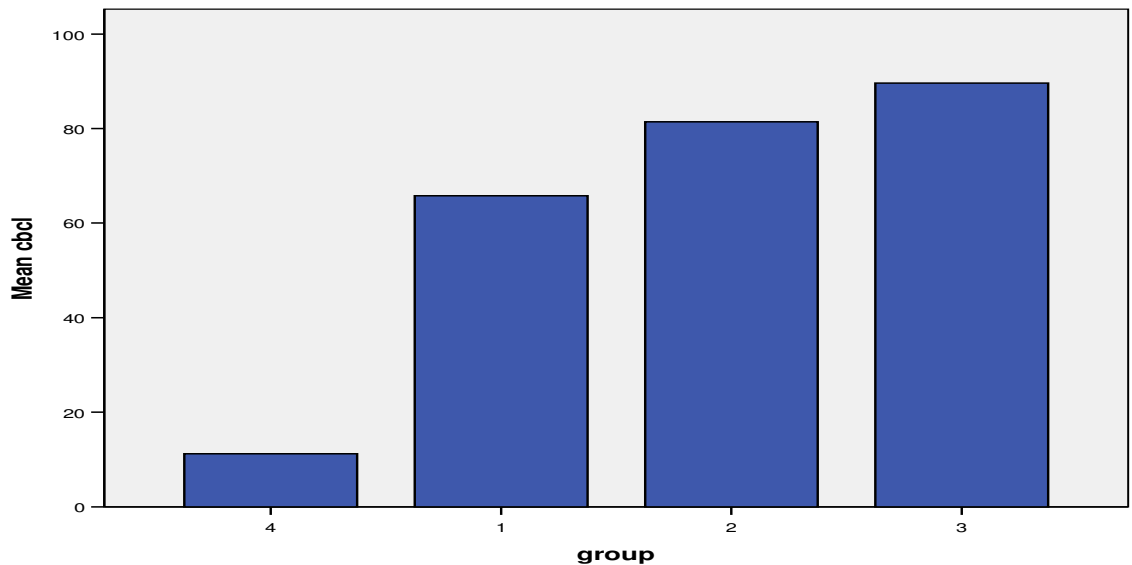
There was a significant effect of group on CBCL ($F(3,88) = 19.08$, $p < 0.01$). Mean scores are shown in Table 3.12 below.

TABLE 3.12 Mean CBCL scores by group

Group	Mean CBCL score	Std Dev	N
1	67.00	33.43	40
2	81.44	29.29	18
3	89.65	49.27	17
4	11.24	10.85	17

There was a significant linear trend ($p < 0.01$) indicating that levels of challenging behaviour increased proportionately with the control group having the lowest scores followed by group 1 then group 2 then group 3 as shown in fig 3.10 below.

FIG 3.10 Challenging Behaviour by group



Planned contrasts showed that the control group had significantly lower levels of challenging behaviour than groups 1-3 ($t(88) = 7.36$, $p < 0.01$), group 1 (non-ASD ASN) had significantly lower levels of challenging behaviour than groups 2 and 3 (ASD with and without diagnosis) ($t(88) = 2.54$, $p < 0.05$), but there was no significant difference between groups 2 and 3 ($t(88) = 0.72$, $p = 0.47$).

CBCL subscales

The CBCL has eight subscales: social withdrawal, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behaviour, and aggressive behaviour. In order to further investigate group differences, mean scores for the eight subscales were explored for groups 1, 2 and 3. Group 4, the control group were not included in this analysis as their scores were significantly lower than the other three groups.

ANOVAs showed no significant differences amongst groups 1, 2 and 3 for 3 of the 8 subscales: somatic complaints; social problems; and aggressive behaviour. There was a significant effect of group on the remaining 5 subscales of: social withdrawal ($F(2,53) = 9.17, p < 0.01$), anxiety/depression ($F(2,53) = 6.21, p < 0.01$), attention problems ($F(2,53) = 4.83, p < 0.05$), thought problems ($F(2,53) = 10.27, p < 0.01$) and delinquent behaviour ($F(2,53) = 5.23, p < 0.01$).

Post hoc tests showed that in the subscale of social withdrawal group 1 scores were significantly lower than group 2 ($p < 0.05$) and group 3 ($p < 0.01$) and similarly in the subscale of attention problems group 1 scores were also significantly lower than group 2 and group 3 (both $p < 0.05$). There was no significant difference in either case between groups 2 and 3.

In the subscale of anxiety/depression, group 1 scores were significantly lower than group 3 ($p < 0.01$) but there was no significant difference between groups 1 and 2 and similarly in the delinquent behaviour subscale, scores for group 1 were significantly lower than for group 3 ($p < 0.01$), and again there was no significant difference between groups 1 and 2 and also no significant difference in either between groups 2 and 3.

However in the subscale of thought problems, group 1 scores were significantly lower than group 2 ($p < 0.01$) and although the differences between groups 1 and 3 and 2 and 3 were not significant, this was the only subscale where group 2 had a markedly higher score than group 3.

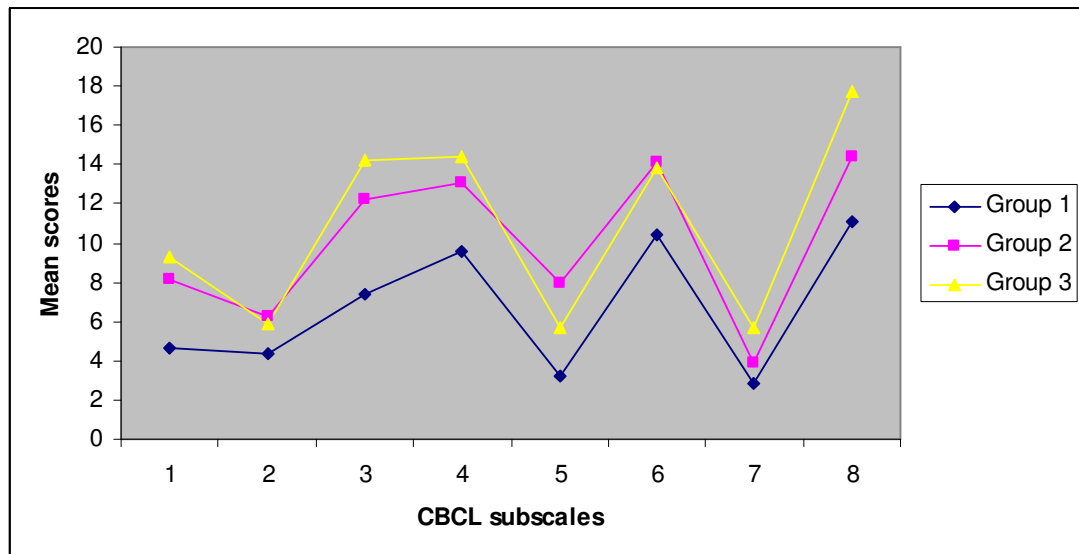
Scores for CBCL subscales for each of the groups are shown in table 3.13 below

TABLE 3.13 95% confidence intervals and standard deviations for CBCL subscale means by group

CBCL subscales	Social withdrawal	Somatic complaints	Anxious/depressed	Social problems	Thought problems	Attention problems	Delinquent behaviour	Aggressive behaviour
Group 1								
Mean	4.64	4.36	7.43	9.57	3.21	10.39	2.82	11.11
95% confidence interval	3.35-5.94	2.78-5.93	4.87-9.98	7.07-12.07	2.12-4.31	8.49-12.30	1.92-3.71	8.15-14.06
Standard deviation	3.34	4.05	6.59	6.44	2.82	4.91	2.31	7.62
Group 2								
Mean	8.14	6.21	12.21	13.07	8.00	14.14	3.93	14.43
95% confidence interval	6.33-9.95	3.28-9.15	8.70-15.72	9.79-16.35	5.88-10.12	12.18-16.10	2.72-5.14	9.32-19.54
Standard deviation	3.13	5.09	6.08	5.68	3.68	3.39	2.09	8.55
Group 3								
Mean	9.29	5.86	14.21	14.43	5.71	13.86	5.64	17.71
95% confidence interval	6.66-11.91	3.22-8.49	10.76-17.67	10.09-18.77	3.55-7.88	11.58-16.13	3.51-7.77	11.29-24.14
Standard deviation	4.55	4.57	5.99	7.51	3.75	3.94	3.69	11.12
Published population normative values								
	2.30	1.20	2.90	1.40	0.40	2.50	1.40	4.80

Fig 3.11 below shows a graph of the pattern of subscale scores for each of the groups. The pink line representing group 2 clearly shows the difference in scoring profile for subscale 5, thought problems. While in general group 1 had the lowest scores, with groups 2 and 3 having similar scores with group 3 tending to score slightly higher, subscale 5 showed a different pattern with group 2 scoring higher than group 3.

FIG 3.11 CBCL subscale scores by group



Subscales: 1 social withdrawal, 2 somatic complaints, 3 anxious/depressed, 4 social problems, 5 thought problems, 6 attention problems, 7 delinquent behaviour, 8 aggressive behaviour

Psychopathology

The subscale of thought problems contains the 8 items: ‘can’t get his/her mind off certain thoughts or obsessions’; ‘deliberately harms him/herself or attempts suicide’; ‘fears certain animals, situations or places other than school’; ‘hears things that aren’t there’; ‘repeats certain acts over and over again, compulsions’; ‘sees things that aren’t there’; ‘strange behaviour’; ‘strange ideas’. These items bear some relation to schizotypal features. The finding that ‘thought problems’ was the only subscale where group 2 (with ASD diagnosis) scored highest and significantly higher than group1, raised the question of whether the additional presence of psychopathological symptoms may be the factor that brings the young person with ASD to the attention of services and makes a diagnosis of ASD more likely.

Previous research has reported that the CBCL subscale ‘thought problems’ could effectively identify ASD in children aged 4-18 (Bolte *et al* 1999). Duarte et al (2003) reported a similar finding for children aged 4-11 and also reported that a factor within the CBCL identified by Rescorla (Rescorla 1988) and named ‘autistic/bizarre’ also identified ASD. The autistic/bizarre subscale was found to be more effective in distinguishing ASD in a group of children with other psychiatric diagnoses while the thought problems subscale was more effective in identifying ASD from a group of schoolchildren. Items included in the autistic/bizarre subscale were: ‘Confused or in a fog’; ‘repeats certain acts over and over again’, ‘compulsions’; ‘strange behaviour’; ‘strange ideas’; ‘withdrawn, doesn’t get involved with others’.

ANOVAs were performed to compare mean scores on the autistic/bizarre subscale for groups 1-3. Results showed that there was a significant effect of group ($F(2,51) = 12.85, p < 0.01$) and post hoc tests showed that scores for groups 2 and 3 were significantly higher than group 1 (both $p < 0.01$). In this study the autistic/bizarre subscale successfully identified those with ASD although it did not distinguish between group 2 and group 3 (those with and without ASD diagnosis). Mean scores are shown in table 3.14 below.

TABLE 3.14 Mean Autistic/bizarre subscale scores by group

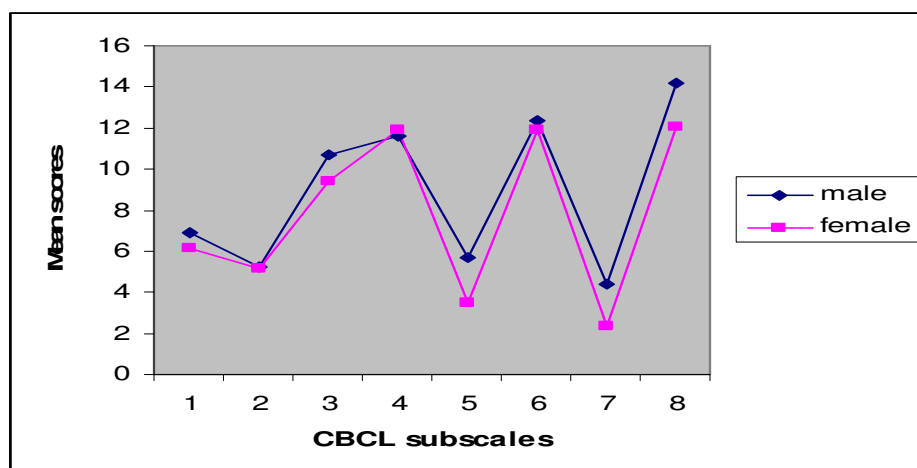
Group	Mean aut/biz score	Std Dev	N
1	6.23	4.26	26
2	13.28	4.94	14
3	12.71	5.94	14

Gender differences

The above findings suggest that a particular behavioural profile involving thought problems make ASD diagnosis more likely for young people scoring in the ASD range. However, there was a high proportion of males (95%) in group 2, (high SCQ score and ASD diagnosis) and it was possible that gender differences could be having an effect on patterns of diagnosis. This could be either because professionals expect that males are more likely to be affected than females or because a particular

profile of behaviour fitting the professional's perception of ASD presentation is more likely to occur in males than females. Investigation of gender differences showed that there was no significant correlation between gender and CBCL total score. Mean scores for males and females across the eight subscales were compared using t-tests. There were no significant differences apart from the subscale of delinquent behaviour where males scored significantly higher than females ($p < 0.05$). Males scored slightly, though not significantly higher than females in total CBCL score and in all subscales except for somatic complaints when scores were almost equal and social problems where female scores were very slightly higher. In the subscale of thought problems, where group differences had been shown, although male scores were higher, the difference was not significant. The profile of male and female scores across the CBCL sub scales are shown in Fig 3.12 below.

FIG 3.12 CBCL subscales by gender



Subscales: 1 social withdrawal, 2 somatic complaints, 3 anxious/depressed, 4 social problems, 5 thought problems, 6 attention problems, 7 delinquent behaviour, 8 aggressive behaviour

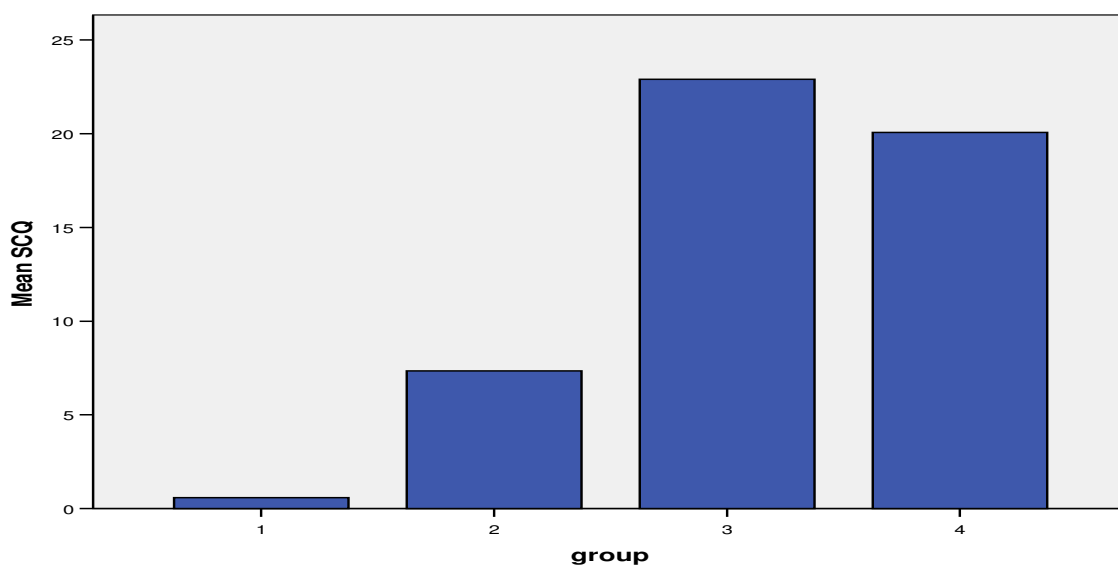
3.8.3 SCQ

There was a significant effect of group on SCQ ($F(3,88) = 135.01$, $p < 0.01$). Mean scores are shown in table 3.15 below.

TABLE 3.15 Mean SCQ scores by group

Group	Mean SCQ	Std Dev	N
1	7.53	3.87	40
2	22.89	5.83	18
3	20.06	3.38	17
4	0.59	0.79	17

In this case it had been hypothesised that group 2 (with an ASD diagnosis) would have higher scores than group 3 (without a diagnosis) and, although the difference between groups 2 and 3 was not significant, planned contrasts confirmed a significant linear trend in the hypothesised direction ($p < 0.01$) as shown in fig 3.13 below.

FIG 3.13 SCQ score by group

Planned contrasts showed that the control group had significantly lower SCQ scores than groups 1-3 ($t(88) = 14.91$, $p < 0.01$), group 1 had significantly lower SCQ scores than groups 2 and 3 ($t(88) = 15.42$, $p < 0.01$), but there was no significant difference between groups 2 and 3 ($t(88) = -1.769$, $p = 0.09$).

3.8.4 Whoqol-bref (Maternal QoL)

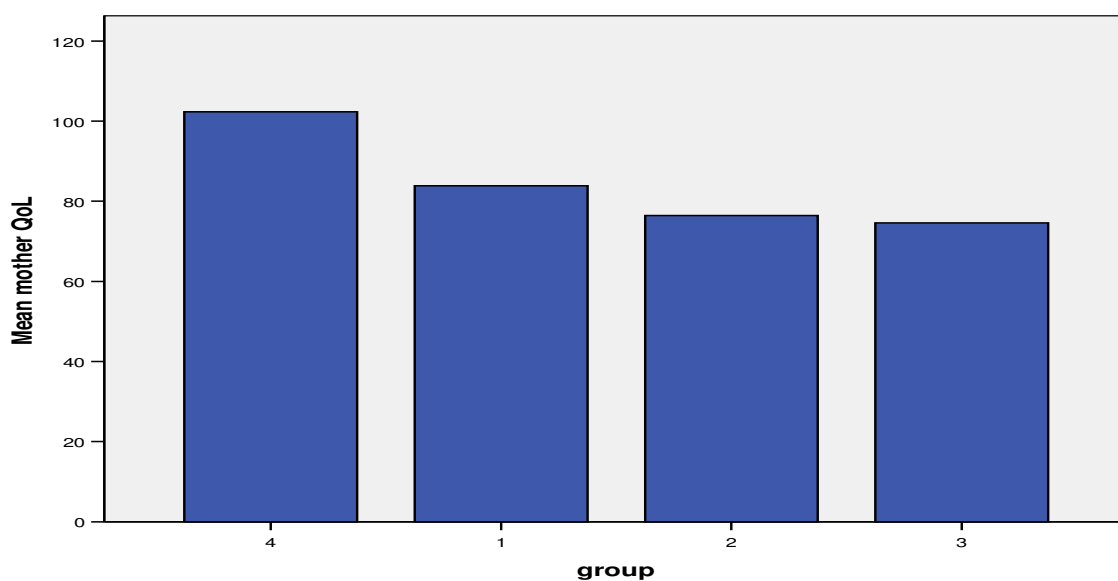
There was a significant effect of group on Whoqol-bref ($F(3,87) = 11.91$, $p < 0.01$). Mean scores are shown in Table 3.16 below

TABLE 3.16 Mean Whoqol-bref scores by group

Group	Mean Whoqol-bref	Std Dev	N
1	84.25	16.15	40
2	76.44	16.46	18
3	74.56	16.56	16
4	102.29	7.52	17

The hypothesised pattern of group scores was confirmed using planned contrasts for linear trend, $p < 0.01$ as shown in Fig 3.14 below

FIG 3.14 Maternal QoL by group



Planned contrasts showed that mothers' individual quality of life was significantly higher for the control group than the other three groups ($t(87) = -5.82$, $p < 0.01$), Group 1 had significantly higher scores than groups 2 and 3 ($t(87) = -2.4$, $p < 0.05$), but there was no significant difference between groups 2 and 3 ($t(87) = -0.36$, $p = 0.72$).

The Whoqol-bref has 4 domains: physical health, psychological, social relations and environment. Further analysis of variance showed that the control group had significantly higher scores than the other three groups in each of the four domains and that there were no significant differences amongst the other three groups in any of the four domains.

3.8.5 FSCI (Maternal stress; groups 1-3)

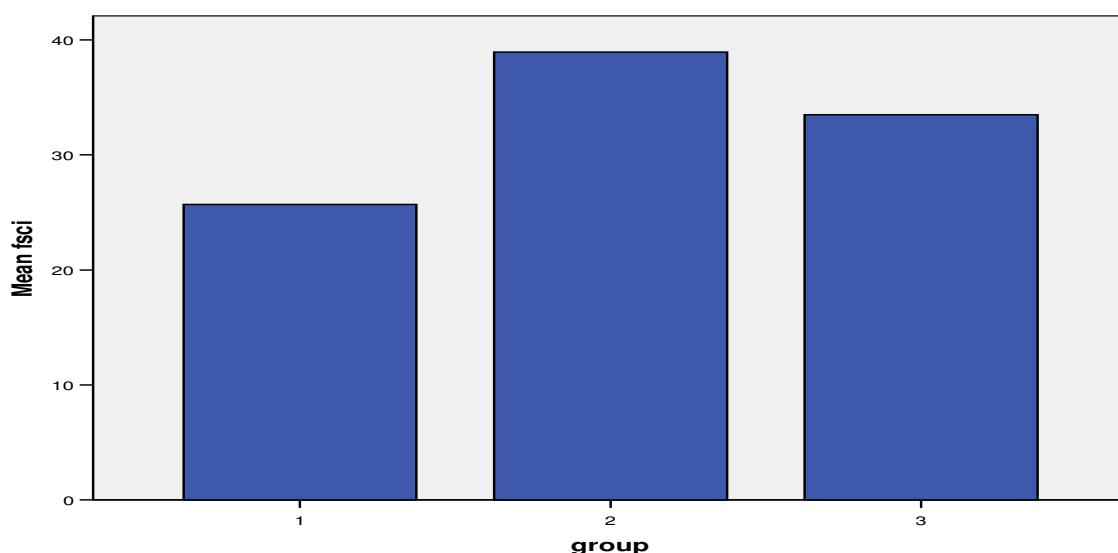
There were only three groups in this analysis as the control group did not complete this measure. There was a significant effect of group on FSCI ($F(2,70) = 4.12$, $p < 0.05$). Mean scores are shown in Table 3.17 below

TABLE 3.17 Mean FSCI scores by group

Group	Mean FSCI	Std Dev	N
1	25.90	15.47	39
2	38.94	17.23	18
3	33.50	18.14	16

No significant linear trend in the hypothesised direction was detected as scores for group 3 were higher than group 2, as shown in Fig 3.15 below.

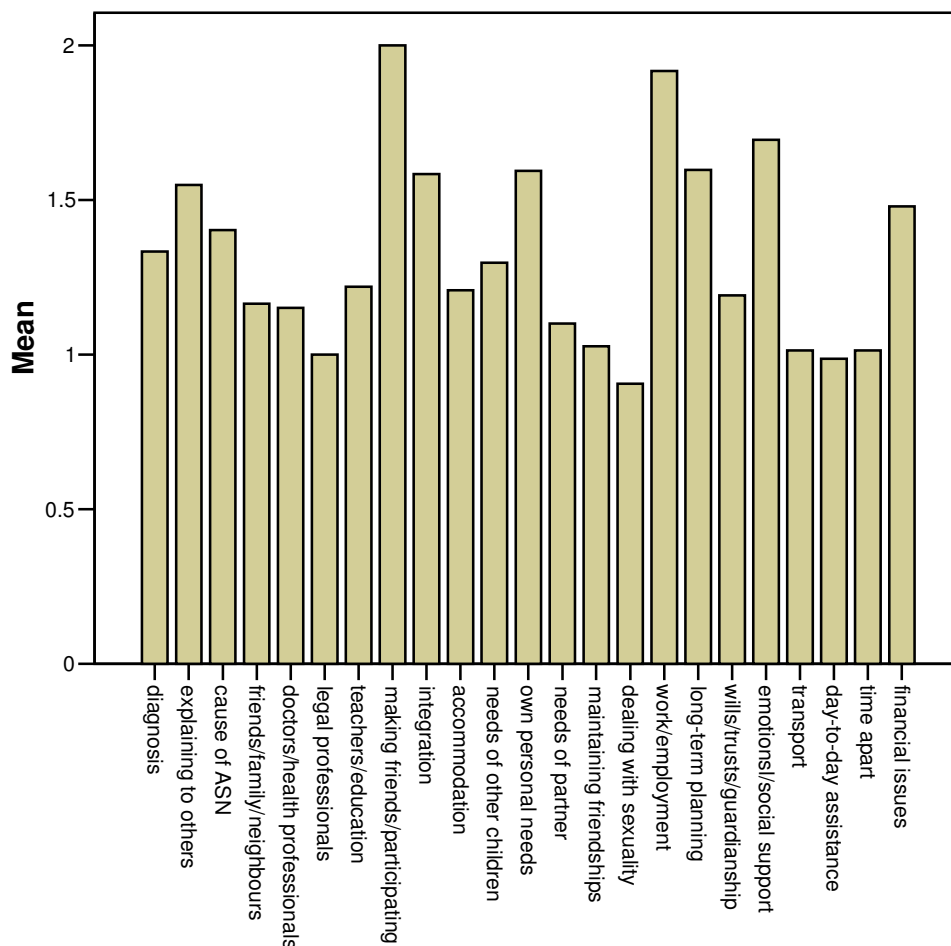
FIG 3.15 Maternal stress by group



Planned contrasts showed that whereas maternal stress was significantly lower for group 1 than group 2 and 3 ($t(70) = 2.74, p < 0.01$), there was no significant difference between group 2 and group 3 ($t(70) = -0.96, p = 0.34$).

Items on the FSCI were each rated by mothers as '0' not stressful, '1' mildly stressful, '2' moderately stressful or '3' extremely stressful. The three items with the highest mean score across all mothers were: 'creating and/or finding opportunities for (the young person) to make friends and participate in activities'; work placements or employment for (the young person' and 'planning for emotional and social support for (the young person)'. Mean stress scores for each item are shown in fig 3.16 below.

FIG 3.16 Mean stress of FSCI items



3.9 Non-parametric analysis

Service Contacts/Usefulness (groups 1-3 only)

For service contacts and service usefulness non-parametric test were used to assess group differences as distribution was found not to be normal.

The Kruskal–Wallis test showed no significant differences between the groups. For service contacts chi square was 1.534, sig 0.465, df(2). For service usefulness chi square was 2.015, sig 0.365, df(2).

Results shown below in Figs 3.17 and 3.18 indicate that although the groups were not significantly different, group 3 had the lowest mean rank scores for number of contacts with services and also for perceived usefulness of services.

FIG 3.17 Service contact by group

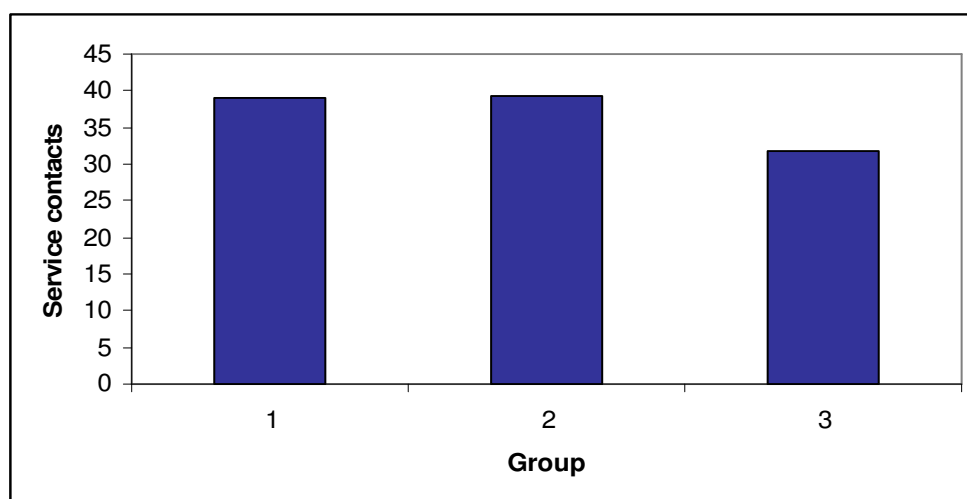
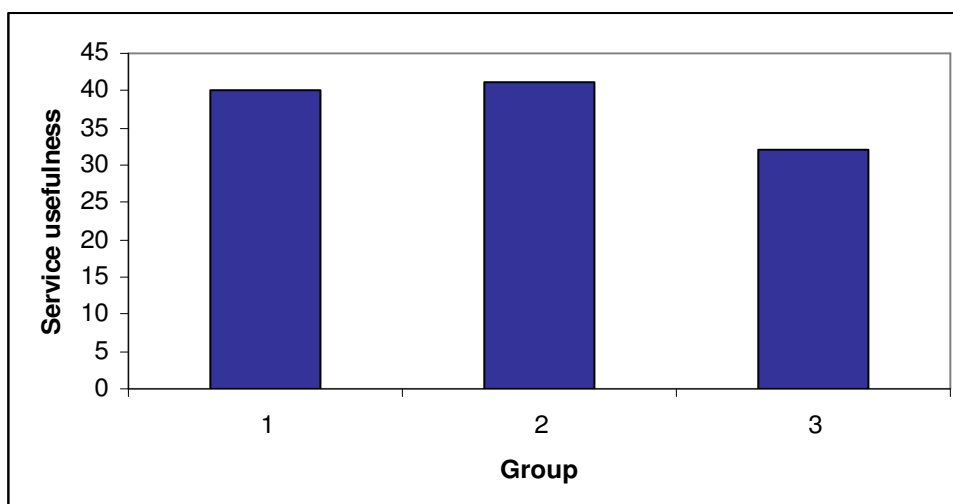


FIG 3.18 Service usefulness by group



Comparison contacts and usefulness of different services

Mothers indicated the number of contacts they had had with each service over the previous month and then rated the usefulness of services as ‘not at all useful’ 0, ‘a little useful’ 1 or ‘extremely useful’ 2. The service that was perceived as most useful and with which parents had most contact was education with a mean score for usefulness of 1.55. The service with the lowest rating of usefulness was social work with a mean score of 0.94 see table 3.18 below.

TABLE 3.18 Comparison of mean service contacts and usefulness

Service	Mean no. contacts	Mean usefulness
Education	1.41	1.55
Social Work	0.35	0.94
GP	0.52	1.06
Community Nurse	0.05	1.33
Other therapist	0.32	1.54
Voluntary Service	0.12	1.50
Other	0.47	1.52

Voluntary services and community nurses were the least-used services and 19 mothers (25%) reported having no contact with any services over the previous month.

To sum up, results from the analysis of variance suggest that mothers of the control group have the highest levels of individual and family QoL and that mothers of young people with non-ASD ASN have higher individual and family QoL and lower levels of maternal stress than mothers of young people with ASD with or without a diagnosis. However there were no significant differences on the main variables between the two groups with positive ASD screening (SCQ) scores.

3.10 Correlations

Correlations were carried out to examine relationships amongst the variables and whether any of these may have been affecting the group differences reported above, and in particular, whether group differences based on the relationships between positive SCQ score, QoL and maternal stress could be being affected by levels of challenging behaviour (CBCL score) which are also associated with high SCQ scores. The control group were excluded from this analysis as their significantly lower scores for level of ASD and challenging behaviour and significantly higher scores for QoL would make it more difficult to detect relationships amongst the other three groups. Pearson's r correlations were carried out amongst all the study variables.

All the main study variables (severity of ASD, maternal stress, maternal QoL, family QoL, level of challenging behaviour) showed significant correlations. Correlations amongst possible confounding variables age, gender and IQ showed no significant relationships with any other variable except for gender and SCQ score. This was inevitable given the expected, and found, preponderance of males in the ASD group. Results are shown in Table 3.19.

TABLE 3.19 Correlations between study variables

	SCQ	FSCI	CBCL	WQB	FQoLS	IQ	Age
FSCI	r(74) = .474**						
CBCL	r(76) = .385**	r(74) = .363**					
WQB	r(75) = -.349**	r(74) = -.564**	r(75) = -.406**				
FQoLS	r(73) = -.378**	r(71) = -.464**	r(73) = -.250**	r(72) = .587**			
IQ	r(65) = -.147	r(64) = -.057	r(65) = -.125	r(65) = .028	r(63) = -.071		
Age	r(76) = .155	r(74) = .157	r(76) = -.034	r(75) = -.065	r(73) = -.204	r(65) = -.40	
Gender	r(76) = -.316**	r(74) = -.067	r(76) = -.00	r(75) = .085	r(73) = .028	r(65) = -.203	r(76) = .001

**p<0.01

3.10.1 Partial correlations

To investigate whether levels of challenging behaviour could be affecting the group differences based on ASD screening score, partial correlations were carried out. As the significantly lower CBCL scores and SCQ scores for the control group were already established, this group was again removed from the partial correlation analysis. Relationships between severity of ASD and maternal stress and also between severity of ASD and family QoL were examined while controlling for challenging behaviour (CBCL score). Results showed that there was still a significant positive correlation between level of ASD and maternal stress and a significant negative correlation between level of ASD and family QoL when level of challenging behaviour was controlled. Although relationships remain significant, significance was reduced in both cases. This suggests that while levels of challenging behaviour are having an effect on maternal stress and on family QoL they do not account for group differences in this study. Partial Correlations controlling for CBCL are shown in Table 3.20 below.

TABLE 3.20 Partial correlations controlling for CBCL

Control variable		FQoLS	SCQ	FSCI
CBCL	FQoL	1.00		
	SCQ	-.319**	1.00	
	FSCI	-.415**	.369**	1.00

**p<0.01

3.10.2 Non-parametric correlations

Correlations between service contacts and usefulness and the other main study variables were carried out using Spearman's correlation coefficient. This showed that service contacts and service usefulness were strongly correlated with each other ($p<0.01$). There was only one other significant correlation: mothers stress (FSCI score) showed a positive correlation with service contacts ($p<0.05$).

3.11 Support for Study Hypotheses

1. Family quality of life (FQoLS score)

The FQoLS satisfaction weighted by importance score would be lowest in group 3, higher in group 2, higher still in group 1 and highest of all in group 4.

- This hypothesis was supported in that scores were significantly higher for the control group (4) than for each of the other three groups. Group 1 scores were significantly higher than group 3. Although differences were not significant between groups 1 and 2 and groups 2 and 3, all mean scores lay in the direction predicted and this was shown to be a significant linear trend.

2. Child behavioural difficulties (CBCL score)

Group 4 would have the lowest scores, group 1 would have higher scores and groups 2 and 3 would have the highest scores with group 2 scoring higher than group 3.

- Again this hypothesis was supported in that group 4 had significantly lower scores than the other 3 groups. However, there was no significant difference between the scores for groups 2 and 3, therefore the hypothesis that group 2 would have higher scores than group 3 was not supported.

3. Presence and severity of ASD (SCQ score)

Group 4 would have the lowest scores, group 1 would have higher scores, groups 2 and 3 would have the highest scores.

- This hypothesis was supported, there was a significant linear trend although groups 2 and 3 were not significantly different from each other.

4. Maternal QoL (Whoqolbref score)

Maternal QoL would be lowest in group 3, higher in group 2, higher still in group 1 and highest of all in group 4.

- This hypothesis was supported in that scores were significantly higher for the control group (4) than in each of the other three groups, with group 1 scores significantly higher than those in group 3. Although differences between groups 1 and 2 and groups 2 and 3 were not significant, all mean scores lay in the direction predicted and this was shown to be a significant linear trend.

5. Maternal stress (FSCI score: groups 1-3 only)

Group 1 would have the lowest scores, group 2 would have higher scores and those in group 3 would have the highest scores.

- This hypothesis was supported in that group 1 had significantly lower scores than group 2 although not than group 3 and there was no significant difference between the scores for groups 2 and 3.

6. Level of service provision (number of services accessed: groups 1-3 only)

Group 1 would have the least contact with services, group 3 would have greater contact with services; and group 2 would have the most contact with services.

- This Hypothesis was not supported. There was no significant difference between the 3 groups in numbers of service contacts.

3.12 Discussion

3.12.1 ASD diagnosis

A main focus of this study was to examine differences between the experiences of mothers and families of young people with ASD diagnosis and a group with possibly undiagnosed ASD, i.e. scoring above the SCQ cut-off for ASD but having no diagnosis. Stresses related to parenting a child with ASD have been well documented as have the benefits reported from having a diagnosis in helping to explain a young person's difficulties. This study investigated whether there were significant differences between the two groups which might explain why some young

people receive a diagnosis and others do not and also whether there were significant differences in outcomes, particularly in terms of maternal stress and family QoL, between the groups scoring above the ASD screening cut-off, with and without a diagnosis.

Although the two groups were found not to differ significantly in terms of severity of ASD or level of challenging behaviour, there was a marked over-representation of males in the group with an ASD diagnosis. This could be due to an expectation amongst professionals that girls are less likely to be affected by ASD or it could be because of differences in the behavioural phenotype of ASD between males and females with more recognition of male ASD-related behaviours.

In terms of challenging behaviour the two groups differed significantly from the control group, although not from each other. The undiagnosed group showed slightly higher levels of challenging behaviour suggesting that this is not the factor that alerts professionals to a possible diagnosis of ASD.

3.12.2 School placement

The young people in group 3, with high SCQ scores but no diagnosis, had the lowest proportion of pupils in mainstream school (19%). Group 2, with high SCQ scores and an ASD diagnosis, had 39% in mainstream and group 1, with ASN but no ASD, had the highest proportion in mainstream (51%). There was no significant difference in IQ between group 1 and group 2, suggesting that the diagnosed presence of ASD made attending special school more likely. However group 3, who had no diagnosis of ASD although scoring above the ASD cut-off, had the highest rates of attendance at special school suggesting that this difference is related to behavioural characteristics of young people with high SCQ scores rather than the presence of an ASD diagnosis. Group 3 also had a lower mean IQ than group 2, although the difference did not reach significance, which could be a further explanation for the increased proportion attending special education. However, this seems unlikely as group 3 did not differ significantly in IQ from group 1 (non-ASD) who had the lowest proportion of participants in special education. This suggests

that although placement in a special school is more common amongst young people with ASD, it does not increase the likelihood of a young person with ASD receiving a diagnosis and is not related to whether such a diagnosis has been given.

3.12.3 Challenging Behaviour

It is possible that differences in levels of challenging behaviour could account for higher levels of special school placement for both the ASD groups, although not for the difference between groups 2 and 3 as they were not significantly different in levels of challenging behaviour. The young people scoring above the ASD cut-off had higher levels of challenging behaviour than the other two groups and this was also demonstrated by significant positive correlations between challenging behaviour (CBCL score) and severity of ASD (SCQ score). There were also significant positive correlations between CBCL score and maternal stress and significant negative correlations between CBCL score and family and maternal individual QoL. These results suggested the possibility that the presence and level of challenging behaviour may be a significant factor in group differences which may have been influencing differences based on ASD screening status. Partial correlations were carried out to look at relationships between SCQ score and family QoL and maternal stress (FSCI) while controlling for the effect of CBCL scores. These showed that although the significance of the correlations were reduced when controlling for CBCL, they remained significant. This suggests that although levels of challenging behaviour have a negative influence on both maternal stress and family QoL, the effect does not account for group differences.

3.12.4 Quality of Life

In terms of family and mothers' individual QoL, results suggest that the presence of a young person with ASN within the family has a detrimental effect and that when the young person also has an ASD or a level of social communication difficulties commensurate with ASD but without a diagnosis the effect is more marked. In this study, mothers in the group without a diagnosis had the lowest levels of family QoL, although this result was not significant, it suggests that quality of life for those

families for whom there is no explanation for the young person's difficulties may be even more adversely affected.

For maternal stress, access to services and satisfaction with services the results followed a similar pattern, although the control group was not included in these comparisons. Maternal stress was higher in the groups scoring above the ASD cut off than those scoring below.

Level of service contacts was shown not to differ across the three groups. Group differences in perceived usefulness of services were not significant although there was a trend towards group 3, the high SCQ group without a diagnosis, perceiving services as being less useful than the other groups, suggesting a higher level of unmet need for this group.

3.12.5 Quality of life of family members

While fathers' and young people's scores correlated significantly with mother's scores, scores for siblings did not show significant correlations with the scores of any other family members. It was difficult to draw conclusions across the four groups as the numbers were too small, particularly for group 3. However, it is notable that the control group with the highest scores for family QoL from the mothers' perspective also had the highest number of other family members completing the FQoL questionnaire and group 3, who had the lowest scores for maternal perception of FQoL, had the lowest number of family members completing questionnaires.

3.12.6 Summary

The results from the quantitative phase of this study suggest that mothers of young people with ASN experience lower individual and family quality of life than mothers of typically developing young people. In addition mothers of young people with diagnosed and undiagnosed ASD suffer more adverse effects than mothers of young people with non-ASN. Although it was predicted that there would be

significant adverse effects for mothers of young people with possible undiagnosed ASD this was not clearly demonstrated in these data.

There were few differences between the two ASD groups suggesting that whether or not the young person receives a diagnosis may be due to external factors such as the area they live in or possibly to maternal characteristics, such as motivation to seek a diagnosis.

The young people in this study came from many different areas and numbers in each group were too small to draw any meaningful conclusions about differences between areas. The qualitative phase of the study, however, described in the following chapter, involved a closer exploration of how these issues were experienced and perceived by mothers and other family members.

CHAPTER FOUR - QUALITATIVE PHASE OF THE STUDY

4.1 Introduction

This chapter describes the qualitative phase of the study which used semi-structured interviews based on the Family Stress and Coping Interview (FSCI) questionnaire (Nachshen 2003) with a subset of mothers to explore in greater depth reported experiences and perceptions of the causes of stress for mothers and within the family. The design of this phase along with the methods of recruitment, data collection and analysis will be described and findings presented.

4.2 Aims

The qualitative phase shared with the quantitative phase, the main aim of the overall study which is to explore the experiences and perceptions of mothers of young people with Additional Support Needs (ASN) and Autistic Spectrum Disorder (ASD). In particular, the interviews were focused on the issues that mothers found most stressful and aimed to identify their perceptions of challenges faced and coping strategies employed. Another aim was to identify mothers' perceptions of how their situation could be improved. The interview questions were designed to provide insight into the processes involved in the mothers' experiences of stress within the family and also to identify positive aspects, by asking them, for example, about successes they have had or by asking for their recommendations for changes in service provision. This represents an empowering approach, aiming to recognise the mothers' skills and knowledge and to acknowledge the value of their experience and judgement (Nachshen 2003).

The qualitative phase was included to complement the findings of the quantitative phase by providing explanations of why certain aspects of mothers' lives had detrimental effects on their levels of stress or QoL and also to highlight possible

protective factors and mothers' perceptions of potential solutions to the challenges they face.

4.3 Method

4.3.1 Data collection

A sub-set of the 76 mothers from groups 1 to 3 (mothers of young people with ASN) who completed the FSCI questionnaire in the quantitative phase of the study were telephoned and then sent a letter inviting them to take part in the interview. An additional consent form was completed for this arm of the study (see appendix 4). The interviews took place at a time and place convenient to the mother, usually in their home. As the interview focused on the experiences and perceptions of mothers of young people with ASN, representatives of the control group were not included in this stage of the research.

Twenty two mothers were interviewed. They were all white, of UK nationality and the biological mother of the young person. Other demographic details are described in table 4.1 below.

TABLE 4.1 Characteristics of mothers interviewed

	N
<i>family</i>	
Single mother	8
Married/living with biological father	12
Married/living with partner (not bio father)	2
Other children in family living at home	16
<i>Mother's Age</i>	
30-40	2
41-50	15
51-60	5
<i>Young person's age</i>	
13-15	6
16-18	11
19-22	5
<i>Area</i>	
Lothian	5
G Glasgow	6
Fife	2
Lanarkshire	4
Renfrewshire	3
Inverclyde	2
<i>Yp diagnosis</i>	
ASD	13
Learning difficulty (only diagnosis)	6
ADHD	3
DAMP*	1

*see appendix 1 for full list of abbreviations

The qualitative section of the FSCI was used as a basis for the semi-structured interviews. This consists of the following five open-ended questions asked in turn in relation to the 3 most stressful items each mother had previously identified in the FSCI questionnaire.

1. What challenges have you faced in this particular issue?
2. What successes have you had and how did you achieve them?
3. How have you tried to cope with this challenge?
Why did you choose this strategy?
Has this been helpful?
4. Has the stress related to this issues changed:
i) over the last 5 years?
ii) in the last year
If changes in stress have occurred, why?
5. What would you like to see change that would help make this experience easier for you and for parents of other children with additional support needs?

4.3.2 Data Analysis

The interviews were audio-taped and transcribed and then analysed using Atlas ti, a qualitative data analysis software package (ATLAS.ti 1993-2007). This software is designed to manage large word-processed data sets and to support a grounded theory type analysis.

The grounded theory approach to qualitative data analysis aims to develop theory which is 'grounded' in the data, by constantly comparing data to test emerging hypotheses. This is known as the constant comparative method (Strauss and Corbin 1998). The Atlas software package allows data to be coded, then organised into categories, which are then grouped together in themes with the aim of finding an over-arching theme or a 'core problem' which is experienced by the participants but has not previously been articulated. This core problem with its core categories must provide a successful explanation of variance within the data. As a theory emerges it

is tested against new data for ability to interpret what is taking place. Theoretical sampling can be used to seek out cases where a particular phenomenon exists to test whether the developing theory has sufficient explanatory power.

The constant comparative method aims to generate categories, sub-categories and codes, to interpret patterns and themes within the data and to ensure rigour by checking and rechecking each emerging code, category or theme against the data and to continue checking data until no new themes emerge. When a code is described the data are searched for other instances of this occurrence as well as instances with a different outcome being expressed. Emerging theory is tested against the data, to see whether it remains true in all circumstances, with the theory adapted as necessary to include an explanation for different outcomes.

In this study analysis involved reading and re-reading the transcripts with relevant quotations from the interviews given an initial coding with additional codes being added as new issues were identified. The codes were merged or divided until all quotations fitted within the coding system. Codes were then grouped in categories and relationships within and across categories were examined. Families of interviews relating to those having a diagnosis of ASD or not and level of service use were also created so that quotations relating to the themes from each of the different groups could be compared.

4.4 Feasibility Study

Before embarking on the full study, data collection from the first 9 mothers was treated as a feasibility study. This set out to ascertain mothers' willingness to be interviewed and the expected length of interviews.

Of the 9 mothers included, all were mothers of young people with ASN. Four were mothers of young people in group 1 (scoring below the cut-off for ASD), 3 were mothers of young people in group 2 (scoring above the cut-off for ASD and having a

diagnosis of ASD) and 2 were mothers of young people in group 3 (scoring above the ASD cut-off but having no ASD diagnosis).

The feasibility study showed that methods were practical and early data analysis produced several possible themes to be explored from the varied and rich data provided by the respondents. The interviews lasted between 40-80 minutes. Mothers had previously completed the Family Stress and Coping Interview (FSCI) questionnaire and interviews were based on the three issues that had been identified as most stressful. If there were not three issues that were clearly the most stressful, the mother was asked to choose which issues she would like to talk about in the interview.

4.4.1 Issues discussed

Mothers chose to talk about a range of issues including: explaining to others about their son or daughter's additional support needs, creating opportunities for their child to take part in social activities, finding college and work placements and meeting their own personal needs. Issues chosen are shown in table 4.2 below.

TABLE 4.2 Issues chosen for discussion by mothers in feasibility study interview

Issue	Mothers choosing
Explaining to others	3
Meeting own personal needs	4
Long-term planning for accommodation	2
Planning emotional and social support	1
Friends and social activities for (young person)	1
Finding work/employment	1
Diagnosis of (young person) as having ASN	1
Dealing with health professionals	1
Meeting the need of other children	1

4.4.2 Conducting Interviews

The instructions for the interview advised asking all the questions in the same order, using exactly the same words to ensure that everyone received the same interview. This was found to be difficult, interfering with the natural flow of conversation. For example, mothers occasionally anticipated a later question in their responses and for the interviewer to then ask a question addressing the same issue again would suggest that the interviewer had not listened to or understood the mother's initial response. This would be likely to discourage the mother from speaking freely and honestly and engaging with the interviewer. On some occasions wording also had to be changed to help the respondent understand.

Although mothers identified the 3 main issues they wanted to talk about, during the course of the interview they often began to talk about other related issues. This was not discouraged as the information was still relevant and helped to identify links between many issues.

One interview was difficult because the respondent had limited understanding and often had no response to questions. This interview was necessarily shorter than the others with the questions presented as simply as possible.

4.4.3 Early findings

Although respondents spoke about a range of issues the following themes emerged as important across the issues and for more than one person:

- Someone to talk to

Most of the respondents said that they would like someone to talk to, often 'someone at the end of a phone', who would appreciate the young person's difficulties and understand the mother's experience.

- Lack of information

Many felt they didn't have enough information. Some were concerned about their son or daughter's diagnosis while others felt that nobody told them about where to get support e.g. financial support or respite care. They had often found information through chance conversations with friends.

- Having to fight for services

Several respondents said that they felt they had had to fight for the services and support their child received.

- Fears for future

Many expressed worries for the future, particularly about what their child would do after school, where they would live and how they would behave and fit into adult society.

- Stigma of special school

Two were worried about this issue and in both cases the young person had been unhappy at attending a special school and blamed the parent for sending them. However, four others were very positive about their son or daughter's experience at a special school.

- Ways of coping

People coped in different ways. Commonly used coping strategies included: seeking to understand and explain behaviour as part of the condition and not as 'badness'; acceptance of the situation and dealing with problems as they arose, and making positive comparisons by recognising that many others face more difficult challenges

4.5 Interviews

A very broad range of issues and ideas were identified through the feasibility study interviews. In order to ensure a broad representation of the sample, to allow comparisons amongst groups and to provide structure, a sampling frame was

developed. This aimed to ensure that interviews included mothers of young people with and without ASD, with and without diagnosis, and also those with a higher and a lower level of contact with services. A further thirteen mothers were interviewed after the feasibility study using the same method as described above.

The sampling frame provided 18 cells to ensure appropriate representation of the range of families and their different circumstances. All the cells of the sampling frame were represented by at least one interview, except for two where there was no study participant meeting the criteria. Four interviews represented mothers of young people scoring above the ASD cut-off, with an ASD diagnosis and with low service use. This was due to an over-representation of this group in the feasibility sample interviewed before the final framework of the sampling frame had evolved. Sampling frame is shown in Table 4.3 below

TABLE 4.3 . Sampling frame for interviews

	High service use	Low service use	No service use
SCQ >21 with diagnosis	1	4	1
SCQ>21 without diagnosis	1	1	0
SCQ15-21 with diagnosis	1	1	2
SCQ 15-21 without diagnosis	1	1	2
SCQ <15 with diagnosis	1	0	1
SCQ<15 without diagnosis	2	1	1

Table 4.4 below shows a simplified sampling frame with each cell filled with the names of the young people in each category whose mothers were interviewed. The young people have been named in order to provide a context for the quotes reported in the findings section.

TABLE 4.4 Young people by group*

	High service use	Low service use	No service use
SCQ +ve with ASD diagnosis (Group 2)	Jack Aimee	Ryan Liam Luke Adam Joe	Logan Lee Aidan
SCQ +ve without ASD diagnosis (Group 3)	Owen Lisa	Brandon Sam	Cameron Nathan
SCQ –ve with ASD diagnosis (Group 1)	Gregor		Daniel
SCQ –ve without ASD diagnosis (Group 1)	Chloe Callum	Jamie	Katie

*All names have been changed with pseudonyms chosen from a list of the 100 most popular names for Scottish boys and girls (General Register Office for Scotland 2006)

The analysis was continued with each new interview being transcribed and entered into the Atlas program. Issues discussed and numbers of mothers choosing each issue for discussion in interview are shown in table 4.5 below.

TABLE 4.5 Issues chosen by mothers for discussion in interview

Issue	Mothers choosing
Explaining to others	7
Meeting own personal needs	7
Long-term planning for accommodation	9
Planning emotional and social support	5
Friends and social activities for (young person)	11
Finding work/employment	10
Diagnosis of (young person) as having ASN	2
Dealing with health professionals	4
Meeting the need of other children	2
Dealing with teachers/education system	5
Dealing with (young person's) sexuality	1
Trusts and guardianships	1

Although the chosen sub-set of issues in table 4.5 above were identified at the start of each interview, during the course of the interview, as had happened in the feasibility study, mothers tended to discuss other issues that were related. This explains why, for example, although only two mothers specifically chose to discuss the issue of the young person's diagnosis, there were several related comments on the transcripts of other mothers' interviews. Although there was a planned structure around the three chosen issues, it was felt useful to allow other issues to be discussed as they arose. Directing the discussion back to the chosen issue may have disrupted the flow of conversation and could have placed a restriction on the mothers, making them feel less free to express their opinions. It was also felt to be important that the mothers had a sense of being asked about issues they felt were relevant for them and in allowing them to express their opinions freely it was hoped to enhance this sense of control over the discussion.

4.5.1 Rigour

In quantitative research reliability and validity are key concepts to establishing the veracity of findings. In qualitative research different criteria are used to achieve these aims: credibility and transferability or generalisability.

Credibility

Credibility refers to whether there is evidence that the information being presented is believable, in that it fits with what is already known and how has the researcher assured this. In this study credibility of the research is addressed in part by the measures described above which ensured that an appropriate sample was recruited. Other measures to ensure rigour included the following:

- During each interview the researcher reflected the respondents' viewpoints back to them to check whether their perspective was being understood or to clarify meanings.
- All interviews were audio-taped and transcribed and entered into the Atlas ti programme ensuring that the actual words spoken were always easily accessible when carrying out analysis.
- During the interviewing phase of the study, the contents of the interviews were discussed with supervisors, both expert in the field, who were able to reflect on the findings and interpretation.
- The published literature was also examined and research in related areas was found to refer to similar experiences amongst parents or carers.

Transferability

Although the sample consisted of those mothers who were willing to complete the interview and may not constitute a truly representative sample, the following measures were included to improve transferability or generalisability of findings:

- Mothers were selected according to a sampling frame based on SCQ score, diagnosis of ASD and level of service provision (see Table 4.3,

p.106). This created a stratified purposive sample in order to illustrate subgroups, facilitate comparisons and ensure broad representation of the range of experiences of mothers in the study.

- The combination with the quantitative phase of the research allowed the evidence, for example of issues relating to mothers' perceived QoL, to be established.
- The sample was based on a theoretical model where the sample group was already known to experience the phenomena in question.
- Mothers in the sample came from a range of backgrounds and geographical areas within Scotland, ensuring that the experiences of a broad range of respondents were included.

4.6 Findings

Quotes from the transcribed interviews were assigned to 82 codes which were then grouped into families of codes or categories, which in turn related to the main themes emerging from the data. Many quotes were assigned to more than one code and there was much overlap amongst the codes and categories but the process of assigning codes to different categories and themes allowed exploration of the relationships amongst the issues arising from the data. A complete list of codes, categories and themes is included in appendix 5.

The four major themes emerging were: coping, disability issues, support needs, and hopes and fears for the future. The themes were related to each other and many codes were assigned to more than one theme. For example, the three issues most commonly chosen for discussion in interview were: creating opportunities for (young person) to make friends and participate in activities; finding work or employment and planning long-term accommodation. Each of these issues was related to the themes of mothers' coping strategies, disability issues, perceived support needs, and also to aspirations as mothers expressed their own and the young person's hopes and fears for the future. Each of the four themes is discussed in turn below.

4.6.1 Coping

Mothers were specifically asked about coping strategies used. Findings were in line with previous research, with parents valuing their own past experience, availability of someone to talk to and often adopting a strategy of trial and error (Grant and Whittell 2000). Several categories were included in the theme of coping and these are described below.

Acceptance

When asked how they coped, mothers found it hard to define coping strategies but often talked of how they had come to accept their situation. Mothers often had a sense of dealing with things because there was no alternative, 'just getting on with it' or 'making the best of it'. For example, Liam's mother said:

You have just got to deal with it you know, that's your life.

In answer to the same question about how she coped, Owen's mother expressed a similar attitude:

I cope to the best of my ability, you have no choice but to cope. You can sit there and say 'Oh well I can't cope' but that's just not what life's about you've got to take life's ups and downs. You've got to make the best of what you've got, you have no choice.

Similarly Sam's mother felt that she accepted the situation and also had a sense of taking control by choosing to have a positive outlook:

You just get on with it. You can say your glass is half full or it's half empty and you decide what it's going to be.

Aimee's mother expressed a sense of her daughter being accepted for who she is by the whole family:

I have never really been one to bleat on about 'poor me, look what I've got, I have got a special needs daughter'. I mean she's ours, we

love her and adore her and we just get on with living life, do you know what I mean?

Taking it as it comes

Associated with acceptance was the notion of ‘taking it as it comes’. In terms of coping mothers often claimed to cope by dealing with each situation as it arose and focusing on the here and now. Liam’s mother said:

I don’t actually have a coping strategy to be honest, you just do it, if it doesn’t work then you just know not to do that again. You deal with the situation as it arises, that is what I do now.

Luke’s mother was not confident that Luke would cope with going away for a weekend with his youth group. Although she was desperate for a break she coped with uncertainty by accepting the situation with realistic expectations and deliberately not anticipating more than ‘one step at a time’:

He has not been once so I mean I am not getting my hopes up, but we will do that step first and then see how he gets on. I mean we might get a phone call at 11 o’clock on Friday night saying can you come and get Luke (laughs).

The Double ABCX model of coping (McCubbin and Patterson 1983) suggests that available coping resources and cognitive appraisal of the situation have an important influence on the ability to cope. In this study cognitive appraisal included making sense of their situation, giving meaning and developing a positive self image which could serve as a protective mechanism. Mothers wanted to understand, to learn and to grow through their experiences; they often described how they had learned to adapt or become wiser and this was associated with seeing themselves as ‘lucky’ or ‘better off than others’ and also with a perception of themselves as a good mother. The mechanism of positive reappraisal refers to the mothers’ attempts to look for a more positive interpretation of their circumstances. This coping strategy has been associated with higher levels of subjective well-being as compared with ‘escape-avoidance’ strategies (Lam *et al* 2003; Glidden *et al* 2006).

Logan's mother had been told for many years that her son 'would catch up' and was devastated when he eventually had a diagnosis of learning disability and ASD confirmed at age 12. She described learning to cope as she began to accept the change in expectations for her son and how this had caused her to reassess her perspective on life as she realised and accepted that that no one can know what might happen in the future to them or their family:

As soon as they diagnosed him, I just went like that, 'he's never gonnae get married, settle doon, he's no gonnae have a normal life, he is never going to have a girl friend, he's never going to make me a granny', you know, these kind of things. And then, as you get older and wiser yourself, you go, 'naebody's got a guarantee anyway'.

Learning to adapt

Learning to adapt was sometimes associated with 'letting go' as the young person grows up, with taking advice from others and learning from experience. It was also often associated with a sense of personal growth in terms of attributes such as maturity, understanding, tolerance, wisdom and having something to offer others who were at an earlier stage or who did not have these strengths. Daniel's mother described how going back to work helped her to develop personally by 'letting go' and finding a better balance in her life:

In going back to some employment it started to take the focus, the main focus away and there was other people being introduced into my own life. I do other support work as well and I gradually became more involved in that as well so that I was starting to develop a balance and started to see that Daniel was a lot more capable than I was allowing him to be.

Katie's mother felt that she had learned, as she grew older, to be more focused and less angry, particularly when dealing with professionals:

I'm much more interested in getting what I want now and if you're angry you immediately create a wall between the professional that you want something from so I'm more focused on getting what I want.

Owen's mother also felt she had become stronger and had developed coping skills. She described the same perception as Katie's mother that anger is a barrier to receiving help from professionals:

Over the years the older the child gets sometimes it gets easier and sometimes it does not. You learn to cope with it, you learn to negotiate and you get a bit stronger. Sometimes you're at the end of your tether. I'm not saying it's easy. You learn to try and sit back and reflect on it and sometimes (in the past) I would just breenge¹ in and shout and everything. Shouting doesn't make it any easier, that makes it worse, if you're calm you're a calm parent and you have the support of people and they'll give you some help.

Nathan's mother felt that she had learned to be more assertive and to challenge professionals:

I used to sit in meetings and it's very.. you know, jargon, you just go home and say 'well what was that about?' But eventually you start getting a bit cheeky, and you say 'well what does that mean?', 'I don't understand that', you think, well if it makes you look silly, it makes me look silly, but I'm not a doctor, I don't know what this particular word means. So I think you just get a thick skin.

Logan's mother also felt she had had to learn to be more assertive in order to meet her son's needs:

I was always very quiet, wouldnae say anything, wouldnae complain whereas now, I'll... you know what I mean, but I felt as if I, as if I had the highest voice because I didnae like what was happening, so you had to stand up.

¹ *BREENGE*, v. *To rush forward recklessly or carelessly; to plunge; to make a violent effort.*

Dictionary of the Scots Language (DSL) <http://www.dsl.ac.uk/dsl/>

Katie's mother described how having a daughter with ASN had forced her to re-evaluate her life as she gained a new focus in working towards addressing the needs of young people with ASN like her own daughter and how she saw this as a positive experience:

My whole emphasis in life changed and I'm really grateful.

Better off than others

Mothers often considered themselves as better off than others. This was often associated with a sense of themselves as being better able to cope because of learned skills or knowledge or because of personal characteristics such as strength or assertiveness or confidence which they perceived some others might not possess. This in turn was often associated with a desire to help and support others and this was recognised by at least one mother as a coping mechanism and perhaps as a way of avoiding personal difficulties by channelling energies into addressing other people's difficulties rather than their own. A sense of being 'lucky' or better off was also often associated with their child's disability being less severe than others.

In terms of comparison with others, Chloe's mother appreciated that although her daughter had difficulties, she compared her situation favourably with parents of children with more severe disabilities or behaviour problems:

There's always people worse than yourself, you know. Like I used to take her to (children's hospital) and see other kids with really bad problems and I know through Chloe's school, Chloe is one of the kids that's no as bad behaved as some of them. Some are worse behaved, and they've got more learning difficulties than Chloe, so I always feel that I am lucky, in a way, because it could be worse.

Owen's mother also compared herself favourably with mothers of more severely disabled children:

I'm probably one of the fortunate ones that my son can walk and talk and feed himself. There's a lot of children not so fortunate. I feel quite humble for the child I have.

Sam's mother had had a difficult pregnancy and felt that Sam's difficulties were milder than she had expected and was grateful for that:

So I could have had a child that was profoundly disabled and actually I could not have a child at all because of what I went through with him. You know they all warned me, at 25 weeks I thought I was having him. So you've got to be grateful for small mercies and say 'well, if that's all that's wrong with him we're doing all right'.

Sam's mother also worked in a secondary school and considered herself to be more well-informed than some other mothers and expressed concern for those she considered less able to cope than herself:

I'm so used to doing things for myself. I know colleges and universities I know how the system works and things and therefore I know who to target and ask and I've got no qualms because of my job as well finding information I know exactly what to do and I just do it and I find it and I don't think about it too much. What worries me is the kids whose parents don't.

She also considered that, unlike herself, other parents may be intimidated by professionals and not have the confidence to challenge them if dissatisfied, as well as not knowing where to turn if they needed help:

....but then I know... lots of parents don't know and that's what really frustrates me. I'm bolshie enough and I don't care who I talk to but there's other parents who think teachers are gods, doctors are gods and educational psychologists even more god-like and I'm like I've got a healthy disrespect for them all, they're only human beings at the end of the day no matter what their title is.

Katie's mother was heavily involved in raising awareness and supporting other parents, she perceived her own strengths as being something she could share with others:

I just set about finding the boundaries myself and finding the answers because we didn't get enough answers I don't know how many parents can do that and I think not very many judging by the phone

calls I get. I think that's one of the driving forces. I know I'm strong and I know how to find information and I am obsessed by protecting other parents so they shouldn't have to feel this.

Katie's mother's perception is supported by comments from other mothers about feeling intimidated in meetings with professionals. Although most mothers tend to describe how they have become stronger because of the perceived need to advocate for their son or daughter, they often describe occasions when this has been difficult. Owen's mother describes a meeting at school:

I feel sometimes very much intimidated by professionals. I feel that if there's say 10 people sitting there: head teachers, principal head teacher, deputy head teacher, psychologist, maybe doctors and maybe have an assessment and you try to speak to them and explain things sitting here with all this paper work highlighting everything that your son has done.

4.6.2 Disability Issues

Many mothers reported that their son or daughter did not wish to attend special groups or activities for people with disabilities but desperately wanted to be part of the mainstream. There was a sense that for this particular group, who generally had mild disabilities that they didn't fit with disabled provision but equally did not quite fit in the mainstream, although this is where they overwhelmingly wanted to be. In some cases the young person had experienced the social stigma of being identified as having a learning difficulty or being different. Many of the young people were very unhappy at having been sent to a special school and often tried to cover this up when mixing with typically developing peers. In other cases young people had not been accepted within a social group and had found this very hard to accept. Nathan's mother told how Nathan used to lie about which school he attended so that other young people in his athletics club would not know that he attended a special school:

He is very conscious of being 'special needs' you know 'special group' you know? He hates that with a vengeance, he absolutely hates that.

Callum's mother also told of the trauma Callum suffered when his friends discovered that he attended a special school. He also had lied for several years about which school he attended and when the truth was discovered he was devastated and felt humiliated. His mother felt that although he had benefited from the supportive environment his school had provided, any benefits were outweighed by his shame at the perceived stigma of attending a special school:

Callum, because his learning difficulties weren't that bad, had they been worse, he wouldn't have been able to have lied so well. The fact that he lied for these years amazed me, and I think what really upset me was the trauma that he must have been going through to continue with this lying all the time, every day and I thought being a teenager was hard enough for him to cope with in his peer group but the fact that his friends found out he was at a special school was just devastating for him.

Lee's mother also found it difficult to find appropriate opportunities for Lee to socialise because his Asperger syndrome (AS) was very mild:

I know there's things for autistic children... Lee is so on the borderline that there are groups for children that have disabilities that are more severe than Lee and there's groups for kids that have no difficulties but for somebody like Lee there's nothing.

Lisa's mother knew that Lisa didn't want to be thought of as 'disabled' and although she found it difficult to make friends, the possibility of attending a social group for young people with disabilities did not seem appropriate:

The difficulty with those sorts of things is...the other youngsters she's going to come in contact with in groups like that have all got problems.... she needs a 'normal' for want of a better word, friend.

Similarly Aimee's mother felt that Aimee didn't quite fit within any of the levels of provision available at her college:

One of the difficulties with College is that it is, you're either in the inclusion or you are in mainstream and there is really nothing in between, and I think Aimee maybe would fit in between, but that's

the difficulty. I mean they have different levels within the inclusion and Aimee is obviously high functioning.

There were contrasts in mothers' responses to their perception of their own son or daughter being less severely or obviously disabled than many other young people. The young people in this study generally had only mild disabilities that would not be at all obvious to others and while, as described above, mothers were often grateful that their child was not severely affected, there was also often a sense that a more severe or obvious disability would be accepted more readily by family, friends, professionals and the public. For example, Sam's mother felt that she had struggled trying to explain Sam's mild learning difficulties and behaviour problems to teachers and while she had already expressed a sense of being 'lucky' that Sam's disabilities were not much more severe and disabling, she said:

Now if he was deaf or blind or in a wheelchair it would probably be easier. Teachers will take it on board this child can't do 'x' but behavioural problems... and there are so many different behavioural problems.

Lisa's mother found that even family did not understand her difficulties, because of the lack of any outward sign:

They don't understand the difficulties that Lisa has because there's nothing physical to see, you know if she was limping around on crutches all the time I think you'd get more sympathy from the family, but if that's what family think then what hope have we got?

Bad behaviour

The notion of people not understanding because there is no visible disability is often related to lack of acceptance by others who dismiss the young person's difficulties as a sign of bad behaviour or poor upbringing. This response from others was reported for family, friends, the wider public and amongst professionals.

Liam's mother describes her experience of trying to explain her son's unusual behaviour to people she meets:

People just don't understand, they look at this normal person and can't understand why he behaves in the way he does. And many, especially older people tend to say 'that boy needs a good slap' and you think, no, he doesn't and you try to say to them 'look he's got Asperger's' and they look at you as if you've got horns. You try to say to them you know 'it's a behaviour, it's not actually a behavioural thing it's emm he doesn't see the world the same way as we do. He doesn't understand situations like we do.' They're not receptive to that you know, to them he's just a bad boy and needs a good skelp².

Brandon's mother had also had difficulty explaining his behaviour particularly to family members, she exemplified their reactions as:

He doesn't look disabled so he shouldn't be doing that, he's a normal person so why is he behaving like that? It's pure bad behaviour.

Gregor's mother also found family as difficult to deal with well as others outside the family:

When people look at Gregor they think there's nothing wrong with him. My brother's the same 'there's nothing wrong with him all he needs is a skelp²,

and Joe's mother reported a similar experience:

It is difficult explaining to other people because he doesn't look like there's anything wrong with him so people think he's just ignorant or bad behaved.

²*SKELP, n. A blow, slap, smack.*

Dictionary of the Scots Language (DSL) <http://www.dsl.ac.uk/dsl/>

Being a good mother

For many mothers the perception of other people is overwhelmingly felt to be that their child is just badly behaved. This may be difficult for mothers to deal with as there is a perceived implication that the behaviour may be due to their failure in parenting. Mothers often spoke about doing their best, trying their hardest and making things as good as possible for their son or daughter, this may be associated with the idea of being a 'good mother' and the desire to be perceived by others as such. Todd and Jones (2003) also reported that mothers felt professionals judged them as incompetent or unworthy. This challenge to mothers' perception of self as being a good mother was particularly hurtful:

Owen's mother felt that she was judged as a 'bad parent' because of lack of understanding of the reasons for her son's difficult behaviour:

....as though we have not brought the children up properly or not good parents it's actually the parents' fault and they believe the children are not brought up in the proper environment and also the children are extremely badly behaved but however that is not the case.

Joe's mother tells a particularly harrowing tale of her experiences when Joe was very young. Joe later received a diagnosis of ASD. Although the fact that he was given medication for his behaviour at the time suggests that the professionals were aware of his problems, Joe's mother still clearly perceived that she was being blamed for his behaviour.

When he started nursery I was told that he was just a misbehaved boy. They put him on Ritalin because nobody would take him and he literally didn't sleep. I used to waken up back of 6 in the morning after maybe having 2 hours sleep he'd be up on top of the wardrobe not a stitch on, he escaped sometimes.. it was absolutely horrendous and I would feel bad as a mother, especially a single mother, because I'd went through a horrific divorce and my other son was a cot death and I used to feel that I was inadequate and I was scared to tell anybody, didn't want them to think I was incapable.

Aidan's mother also felt that teachers linked Aidan's bad behaviour at school with her failure as a parent. She felt blamed and judged for his behaviour and found reassurance when she met other mothers who were having similar experiences:

You think that your child is the only child in the school that is causing bother, but that's how the teachers make you feel. But I said 'well, at least, thank goodness, I am not the only one it's happening to', because I felt as though I was the worst mother in the world, and I was trying my best to be a good mother. (laughs)

Most of the mothers who spoke about difficulties in explaining the young person's behaviour to other people were mothers of young people who scored above the ASD cut-off. Mothers of young people scoring below the ASD cut-off did not describe such difficulties and in a particular contrast, Callum's mother said that it was not at all difficult explaining to other adults about Callum's difficulties:

Adults do understand quite easily, I mean I don't have a problem at all explaining to adults, in fact I don't really tell people until they ask, I am not going to go about advertising that Callum is at a special needs school. People are quite surprised, and sometimes (say) "Oh really, I wouldn't have known".

Even when people express surprise this is interpreted in a positive way as an indicator that Callum is very well accepted and does not appear different from other young people. This is also a marked contrast to the situation where mothers feel that people do not believe that their son or daughter has ASN. This suggests one possible explanation for the increased stress and lower family and individual QoL experienced by mothers of young people with ASD. It is possible that young people with ASN who do not have ASD behave in a more socially acceptable way and therefore do not face the same difficulties in being accepted within the community or draw the same attention from others as those with ASD. It is also possible that having a diagnosis may make it easier for mothers to explain unusual behaviour and yet the experiences reported by mothers that this is not always the case may explain why no significant differences were found in stress or QoL between the two groups scoring above the ASD cut-off with and without a diagnosis. In addition, the difficulties mothers of young people with a diagnosis report in dealing with differing

professional opinions, with on-going assessments and with vague diagnoses may affect stress and family QoL. The lack of support available after diagnosis for some young people may also explain why having the diagnosis may not necessarily reduce stress or improve QoL.

Making Sense

In spite of difficulties related to getting a diagnosis of ASD, many mothers perceived that there had been changes in recent years and that there was now more public awareness of ASD and AS. Having a diagnosis sometimes helped mothers explain their son or daughter's behaviour to others. Liam's mother, for example, described different responses to Liam's unusual behaviour. Although, as described earlier, people did not always accept her explanation of AS, on other occasions people were more receptive and she felt that this situation was getting easier as public awareness had increased in recent years:

People are talking about it more now, so people are more accepting of what you're telling them. Like one time when I said 'I'm really sorry.. Liam's behaviour... he's got Asperger's' and I was really taken aback because somebody said to me 'Oh, right that's on the autism line isn't it?' and I was like that 'Oh, somebody actually knows' and I said 'uhuh, I'm really sorry' and (they said) 'don't worry about it'.

Having a diagnosis of ASD sometimes gave families access to services and to support groups such as the National Autistic Society as well as the benefit of realising that other people are having similar experiences and that the difficulties are acknowledged as being part of a recognised condition. This may have helped mothers preserve their own sense of being a good mother in that it provided a reason for the difficulties and therefore reassurance that they were not to blame.

Receiving a diagnosis was not always associated with positive experiences however. Many mothers reported that even after receiving a diagnosis no additional help, support or information was made available to them and that professionals and members of the community still generally did not understand the implications of the diagnosis. Aidan's mother had struggled to explain Aidan's behaviour all through

primary school and although he was given a diagnosis of AS at age thirteen, this did not seem to help the situation:

Even in his secondary school, they knew that he had the Asperger's and they knew that he was dyspraxic and that he had got this diagnosis, it didn't change much anyway.

Jack's mother also felt that it had been a long hard struggle to get a diagnosis of AS for Jack and although having the diagnosis helped her to understand and make sense of Jack's behaviour, she felt that no support had been available for her family:

He was given a diagnosis but they are not then able to offer anything else, any other ways of helping or dealing with Jack's behaviour.

Adam received a diagnosis of 'autistic tendencies' at age twelve but his mother also felt unsupported after the diagnosis:

When they did diagnose it, if they'd told me there was help to be had with the autism and how to go about... it would have been ideal. But because I never got told anything and I didn't get any help you know that was ...quite wrong. I feel that I should have been offered some form of help.

Adam's mother also expressed dissatisfaction with the vague diagnosis of 'autistic tendencies'. This supported published findings that unclear or ambiguous diagnoses were a source of dissatisfaction amongst parents of children with ASD (Brogan and Knussen 2003):

It took a few years for the diagnosis and then when I finally got it, all I was told was, 'yes, he has autistic tendencies' and that was it. No follow up, nothing. So I'm just left to think 'well, he's autistic, but to what degree? What does it mean? How does it affect him?'. I never got any information.

Another negative aspect was that some mothers found it hard to come to terms with the diagnosis either because they had previously been given different information or because their child did not fit the image they had of someone with ASD. Logan's

mother described being distraught when Logan was given the diagnosis of AS at age eleven. When asked if receiving the diagnosis had increased her stress, she said:

Only because they had already said he never had it. We were prepared for it the first time, but when he says to us 'naw, go away' we were, 'Thank God it's something he will grow out of'. I think that's how I took it so hard. I think that's how everybody did.

Aidan's mother had searched for a diagnosis, knowing that something was unusual in Aidan's development and looking for an explanation. However, she was surprised when a school doctor suggested that Aidan, then aged eleven, might have an ASD. She describes the different opinions of professionals she came into contact with:

I said 'I have had him at clinics and doctors and nobody ever suggested that.' I had heard of autism, but I always thought that autism was, they didn't speak. Aidan spoke so I was absolutely... She said, 'No, I mean very, very mild' and I think it was her that brought up the Asperger's and when I went back to the Psychologist, she said, no he didn't have Asperger's, but (laughs) when he went to the Speech and Language Therapist through the Doctor in the school, they said, 'Yes, he definitely has, in our opinion.'

Daniel's mother also found the diagnosis of AS, at age 14, hard to accept and the professionals confirmed that there was a degree of uncertainty about the diagnosis:

When he was diagnosed with Asperger's I didn't entirely agree with it, I felt a bit angry, 'he's not Asperger's'. Daniel to me didn't fit the bill. But what they said to me was, 'we know he doesn't fit all the bill but that's the nearest because his original diagnosis was speech and language, round about 9 or 10, but there's something else going on here there's some communication disorder and it took us 4 years because Daniel had to develop and we just had to kind of watch him, monitor him, see what way he was going'.

Gregor, now aged 16, had received a diagnosis of AS at age 12 and although his mother valued the diagnosis as a way of understanding his behaviour and helping to try and explain it to others, she was now waiting to take Gregor to be reassessed:

We've got to go through the whole process again with the Asperger's because the doctor that diagnosed him retired and didn't put it in black and white so we're just waiting to go to get it all done again.

Of the 12 mothers of young people with an ASD diagnosis who were interviewed, 10 described issues relating to the diagnosis of ASD. Eight mothers described having to wait many years to get the diagnosis and many were given conflicting explanations at different times by various professionals. This may reflect the fact that the young people in the study were not severely autistic with the easily recognisable features of classic autism. Their difficulties were more subtle. Six were eventually given a diagnosis of AS, one of autistic tendencies, one of autism. Prevalence estimates for ASD vary considerably (Fombonne 1999; de Bildt *et al* 2005) and diagnostic criteria have changed as awareness has grown of milder variants of ASD (Filipek *et al* 1999; Ghaziuddin and Mountain-Kimchi 2004). This may explain why these mothers and young people, with generally mild ASD or AS, have struggled to get a diagnosis, but it does not detract from their perceptions of lack of support from professionals.

Of the mothers of young people with high ASD screening scores but no diagnosis of ASD only one talked about her son's diagnosis and expressed frustration at the sense of not knowing or not being able to explain his difficulties. Brandon had been given a possible diagnosis of ADHD:

It is just one constant battle because social workers, community workers, doctors like to have a label attached to things and if you've not got that label to say what's the matter with your son they don't listen. We've been told so far that he has ADHD but there's been no actual test done to see exactly what's the matter with him. Can we get something done to tell us, 'Yes your son's this, your son's that'? They don't know. Nobody knows. He's got learning difficulties, 'Well to what extent?' I don't know. I know he's got learning difficulties I know he had to attend special school and they say to you 'yeah, but to what extent?' and you can't tell them because you're not told.

Even the mothers of young people with ASN and no ASD mentioned difficulties relating to not having a clear diagnosis to explain their son or daughter's difficulties.

Chloe's mother talked about not being given any explanation for physical anomalies that were noted and also about wondering whether Chloe could be autistic:

There should be somebody there that you could say "My daughter's got learning difficulties and I'd like to find out exactly what is wrong with her", because nobody's ever said to me Chloe's got..... I always thought she was a wee bit autistic at first, but naebody has ever said to me this is what it is. I know lots of things can happen to kids, but they should be able to tell you, like she went for x-rays for her hands, her's go that way (gestures) and her feet as well and she got them x-rayed, but they didn't tell you anything, they didn't say what's caused this, or lots of kids with learning difficulties have got this.

Both Brandon's and Chloe's mothers talked about 'not being told' about what their children's problems were. This suggests dissatisfaction with the support they have received from professionals in that they perceive that the professionals do have these answers but have chosen not to pass on this information to the parents.

4.6.3 Support

Family

Family was the most commonly reported source of support. Many mothers described how help from family, usually their own parents or siblings, especially sisters had been invaluable. Those who did not have the support they needed often perceived that this was because they had no close family or family members lived too far away.

Although family was the most useful support, relationships within families were not always supportive. There was often a sense that family members did not understand or appreciate their difficulties, were not accepting of the young person and were inclined to criticise the mother's way of managing situations.

Friends/informal support

Informal support from friends was also important and this included those who were also parents of young people with ASN and those who were not. Many mothers told how they found information by word of mouth from friends or other parents or from internet sites rather than from professionals. There was a sense of luck or chance as to whether appropriate services were received or available. Many mothers had been involved with support groups, some had attended and some had been involved in setting them up. However others had tried them and not found the experience helpful or did not wish to be involved because either they did not feel they needed them or they did not value the kind of support they felt the group could offer.

Professionals

There were mixed and conflicting views of support from the various professionals that the mothers had contact with. In some cases professional support was cited as the key to successful adaptation, with many mothers receiving valuable advice and support from teachers, doctors, social workers and others. At the same time there were many reports of inadequate support that left parents feeling isolated, frustrated and angry.

Education

In the earlier questionnaire about services and support, education was named as the service with which the mothers had the most contact and which was felt to be the most useful. In the stress questionnaire, although mentioned frequently, dealings with teachers and the education system were generally considered to be mildly rather than moderately or severely stressful. In interviews four of the twelve mothers of young people with an ASD diagnosis chose to talk about education but only one mother of those without a diagnosis of ASD chose education as a stressful issue that they wanted to discuss.

In general parents had more positive impressions of primary school, feeling that things had been easier at that stage. This may reflect some of the difficulties young people face as they are growing up in terms of behaviour and discipline and the

more apparent deficits in academic ability as young people move towards exams and the increased academic demands later in secondary school.

There were contrasts in opinions about support from school with some mothers feeling that the school did not communicate adequately with them, while another described similar amounts of contact but felt that this was perhaps more than she needed. For example, Owen's mother said:

There's no communication whatsoever. There's no communication, I feel there's very, very little you've only got a parents' meeting maybe once every 6 months or you've got another review meeting twice a year. I feel they should keep parents more informed what's going on, send parent bulletins out, mail shots, education should have more meetings with parents.

However in contrast Luke's mother said:

We have got to make decisions now really, because we have got to put things in place because they take that long to happen. So now we have these future needs meetings. Gosh! I think they come up every four or five months. I'm going 'we've just had one of these haven't we? And then we get another one' (laughs).

These two contrasting perceptions demonstrate the individual differences in mothers' need for support.

Lack of provision

Mothers perceived that there was a lack of provision in many areas of their sons' and daughters' lives. Mothers mentioned lack of college and work placements, accommodation options, respite provision and supports such as befriending schemes. Social workers in particular were seen as being not available, not returning phone calls or following up on agreed plans of action.

Several mothers also felt that they had been discriminated against by services in that the young person was not seen as a priority because the family was there and considered able to provide the necessary support. Ryan's mother considered that

Ryan was not likely to receive help from the over-stretched local social work department:

He is not a priority. There are no social work resources or they are limited. Ryan is not a priority for social work, he is with parents who are willing to look after him, he is safe, he is secure here.

Luke's mother waited a year after discussions with social workers assessing the family's need for respite care. When she eventually contacted them to check on progress, she was told that they would not be able to help her because her family was not considered a priority. Not only was she disappointed at not receiving the help she had been hoping for, but she was angry that the social workers had not contacted her for a year to tell her their decision:

I said 'I have been on this list for over a year and you promised me that something was going to be happening'. He said 'well I thought...', he just passed the buck to be quite honest, he said 'well I only come and do the assessment and I pass it on'. I said, 'yes well why did somebody not tell me that, I have sat waiting a whole year thinking that it's going to be happening, it should be happening soon, somebody should be coming to me, we will be going for visits to... and now you are telling me that I am not a top priority. So I am just no further forward after sitting waiting a year, a whole year'. (laughs). I didn't have a good day that day,

Luke's mother decided that desperate measures would be required before she would get help with Luke:

I think I have got to march him into the office by the scruff of the neck and say, 'Here, there you are, you do something with Luke, because I'm ready to shoot myself' (laughs). Sometimes that's what you have got to do. It's got to be a crisis I think, before they find a place.

Brandon's mother also felt that professionals were unwilling to help because the family was considered able to cope:

They come into the house and they're like, 'Well you've got your mother, you've got your father, you've got brothers, you've got all

that, you don't need the help.' You don't need nothing because you can depend on them to do it.

Although there was a degree of sympathy for social workers in that they were perceived as being busy and over-worked, mothers felt they had no professional to turn to for information or support. Liam's mother had little support from her social worker:

We have only got a part-timer, so she is only in two days a week. So if we don't get her on a Tuesday and a Wednesday, then that's you till the following week and sometimes if you leave messages you know they build up so you have really got to wait your turn.

She felt there was no one she could turn to for advice or information or for support in difficult times:

I would like.... even somebody else who understands the child's condition or whatever that you can pick up the phone and say 'Oh Lord, he's just done...and can you give me any idea what to do?'. Because I feel just now that we are out on a limb.

Lisa's mother also felt that she didn't know how or where to find information about any help or services that might be available:

There was no independent information coming to us, it's word of mouth or something that happens to come along. It's all a hit or a miss. There's no sort of central place you can go and sit down and say 'my kid's got difficulties. This is what they've got a problem with', you know 'What do we do?' And that there would be somebody who could say what all the various services are.

There was also a perception that even if help was available in one area there was little co-ordination amongst the various service providers. In answer to the question 'what would you like to see changed that would help you and your son?' Ryan's mother's reply described the situation where the young person would have an allocated key worker to oversee their contact with other services:

What I think would be really good for kids like Ryan, is to have somebody who deals from the bottom up basically and is the link to all the different agencies that need to be involved with a young person coming out of College or you know, with no future basically, with nothing else planned. You've got supported employment, you've got... and all these people, they work in tandem with each other, but they also work very separately from each other and maybe just somebody, a contact who you trust to deal with benefits, employment, training opportunities but very holistic you know. If say Ryan needs company then there is a group that somebody could find you know.

What Ryan's mother described was very much in line with the recommendations of the Same as You Review (Scottish Executive 2000a) that Local Area Co-ordinators (LAC) should be appointed. The LAC would be responsible for a number of families in their geographical area. Their role would be to support the family and to liaise with appropriate services. This policy is now being implemented across Scotland, although it is clearly not yet reaching all families who would benefit.

Not all contact with professionals was viewed in a negative light and many positive experiences were described. Although mothers had criticisms of organisations and the way services were set up, as well as particular individuals with whom they had had poor relationships, experiences which had a beneficial outcome for the mother or the young person were often described in terms of the successful relationship with the individual professional involved. Good relationships were characterised by a sense that the professional was committed to helping the family, that they listened to the mother or the young person and responded to challenges by giving good advice, organising practical help or by understanding and not denying mothers' experiences.

Luke's mother described how the psychologist had visited the family regularly over a year after Luke received a diagnosis of AS. He gave practical advice about dealing with Luke's behaviour and also spent some time explaining the implications of AS:

He made me understand that Luke couldn't understand our way of thinking, but we.....could try and understand how Luke saw things. So I suppose it taught me to be a bit more tolerant.....definitely the psychologist has helped me loads. I'd say he was worth his weight in

gold because after that I saw the light and I saw things totally different and I could cope different.

Logan's mother described the help and commitment of teachers at Logan's primary school who had responded positively to Logan's diagnosis of ASD:

She (the Headmistress) went to find out about the communication part of it, she went away in her own time and done all that. There was another teacher in the school as well that, she had prior knowledge of it and she especially asked to take the class that Logan was in because she felt that she could... so I mean I was really lucky, we couldnae have wished for better.

Sam's mother described poor relationships with some teachers whom she perceived to be unwilling to listen to a parent's point of view. In contrast, she describes the useful discussions she had with a teacher whose support she valued:

She treated me as an equal and listened to what I was saying and took on board the things I was saying and she would try things. I would suggest things and she would try them and she would suggest things to me and I would agree or disagree or say why and so we did have a good relationship.

4.6.4 Hopes and fears for the future

Many issues raised fears for the future for mothers. These included practical concerns about work and employment, college places, accommodation and financial security. Mothers also expressed concerns over whether the young person would be emotionally supported, be secure and protected and not be prey to those who would seek to take advantage of their vulnerability. They hoped that their son or daughter would be happy and accepted in the community. Mothers wanted to see the young people growing up, becoming independent and making their way in the world. Finding work and being materially secure were important practical concerns for mothers but being happy and fulfilled, achieving their potential, having friends and being accepted within the community were just as important concerns related to the young person's social and emotional well-being.

There was a sense of frustration at the lack of perceived opportunities for employment and accommodation and a feeling that the young person faced many barriers, including stigma and lack of awareness of employers and other members of the public of their potential.

Work/employment

One of the items identified by the FSCI questionnaire as most stressful was 'finding work placements/employment'. This was also one of the issues most commonly chosen by mothers to discuss during the interview. There was a perceived gap in provision after school and most of the young people were at college already or planned to take a college course. This was often seen by mothers as the only option although some felt that their son or daughter would rather get a job than go to college. Lee's mother felt that Lee would prefer to work than go to college:

When they are leaving school, you would like to know all the options, most of the kids are getting offered to go to College, and this is not what a lot of kids want to do.

Adam's mother also felt that college courses had little to offer. Adam had tried a college course but his mother felt that the course had simply repeated what had been covered at school and that he had not enjoyed the course and had gained little from attending. She felt that offering college courses was an easier option for schools than finding suitable work:

The school seemed more concerned with the likes of this college course rather than getting them a job.

Even after the young people had been to college, job opportunities were seen as scarce and support in finding jobs not always effective. Ryan had already completed his college course and had a job coach to help him find work and to support him in any employment, but his future still felt uncertain to his mother:

I suppose it really hits home to you, see when they leave the last place of education, and there's, you run into the brick wall that really becomes stressful..... it's number one I think for us on the list in

terms of stress now, we really want Ryan to be successful in a job and its what he aspires to.

Although Brandon had left school and was attending a college course, his mother was concerned about lack of work opportunities for him after finishing college. She again echoed the need for advice about these issues:

There's no-one really to sit down and say 'right Brandon has these disabilities, Brandon has these problems, they have to be worked on in a work situation' because all we can see ahead of us is Brandon stuck in the house.

Liam was due to leave school soon, he did not have a work or college placement and his mother was also concerned that he would be at home with her every day:

If he doesn't get into that (college course) he's just going to stay in the house with me and I'm like that, 'Oh no you're not because you either have to find a job or you have to go to college or you have to get out at least a few days a week.' I couldn't have him in here that would really suffocate me that would really feel em

Mothers were also very aware of lack of opportunities for suitable employment. Adam's mother worried about finding a suitable career for him:

Because of his capabilities or lack of them, it's going to be hard to get him into a sort of job that he's going to get a career in, like an apprenticeship of sorts. I know that they've got a quota and they've got to take children with disabilities or whatever, but I think it's very difficult.

Ryan had a job coach who worked with him to try and find work placements. His mother recognised the difficulties in raising awareness amongst employers to try and create more opportunities:

None of this works unless employers are really engaged with it, and I think the big thing that there really needs to be more of I think is in engagement with employers.....there needs to be more opportunities there are some excellent companies like (major supermarket chain) that take these kids on and give them a chance, but there are thousands of small and medium sized companies that I

feel if they could get appropriate support and understanding maybe we just don't take the message out enough.

The frustration at lack of opportunities was linked to a sense that the young people were not being enabled to fulfil their potential. For example Katie's mother worked for an organisation that was involved in raising awareness amongst employers about ways they could accommodate people with ASN within their workforce and the possible benefits they could gain from doing so. She described her view of what is needed:

Certain jobs are absolutely made for someone with Asperger's. So sorting post, sitting in front of little boxes with little writing with postal codes and sorting for ages - that suits someone (with Asperger's). Someone with Asperger's often has that special talent so put them to do that. I could never do it in a million years. So to change and accommodate instead of shutting people like Katie and others out, society needs to accommodate and they will be fantastic contributors because they have special talents.

In an example of the same kind of initiative Logan's mother described a successful supported work placement that Logan had undertaken at a local factory. Logan had AS and the job suited his personality and abilities very well. It was repetitive and required consistent accuracy although was not academically demanding and Logan achieved great satisfaction from doing the job well. Unfortunately the factory had now closed and so there would be no opportunity of Logan finding permanent work there:

He did a lot of different jobs, but one of the things was to put 10 screws in a wee bag all day long. Logan loved it. And he was so particular about it, he loved it, he done that and passed it on, done it and passed it on, and you get so many in a box and he took the box to be checked. So it was something he could (do), he picked it up like that, he could do it and he likes everything to be the same, so it was ideal for him.

Cameron's mother was frustrated at the lack of opportunities for part-time work of the kind that typically developing young people might find while at college. She also felt that Cameron and other young people like him had a lot to offer an employer but were not given the opportunity:

They'd be great because they would turn up they'd be reliable they would do as they were told they'd be dead keen

Long term accommodation and financial security

Young people's long term security was a concern for parents, especially when they were no longer able to care for and support their son or daughter. Many had already begun to make enquiries about accommodation but like other services it was difficult to get information and provision seemed to be inadequate. Associated with concerns about where the young person would live were concerns about how they would be able to support themselves. Parents who were themselves financially secure worried about who would protect their son or daughters interests once they had inherited the family's assets and there was a concern that someone could take advantage of their vulnerability.

As the following two comments from Luke's mother illustrate, although siblings had usually been involved in discussions about future plans, mothers generally didn't want to pass on full responsibility to them for their brother or sister. However, most expected that the sibling would oversee the care of their brother or sister to a degree and siblings had often expressed a willingness to do so:

Luckily he's not an only child, there are two siblings there. I'd like to think that they will look out for him, I am sure they will, but he still needs somewhere, he still needs his life, his permanent place, he still needs something and I don't know where that will be.

I don't think (the siblings) should have the responsibility of Luke permanently. I mean, yes, I want them to have him at Christmas and birthdays and in the holidays, and have him stay in their families just to make sure they look out for him.

Similarly Lisa's mum felt that she had to explain to her younger brother that he would have to be prepared to take some responsibility Lisa in the future:

I have to make him start to realise his responsibilities in that department so that it doesn't suddenly hit him when he's in his

twenties or something like ‘crikey, am I going to have to...?’ I’ve said to him ‘you’ll always have to look out for her, it doesn’t matter whether you fall out or whatever you are going to have to be the one to make the first move back in there and keep in touch with her and make sure she’s okay. I’m not expecting you to have to take her into your home or anything like that but you are always going to have to look out for her.’

Friends and a social activities

‘Creating and/or finding opportunities for the young person to make friends and participate in activities’ was scored as the most stressful item of the FSCI questionnaire. It was also the issue most often chosen by mothers to discuss in interviews. In most cases making friends had been difficult for these young people, irrespective of whether or not they had ASD. Many mothers reported that the young person didn’t have any friends, Joe, for example, had real difficulties in finding friends and socialising and his mother described a recent evening when a social worker had taken him out with a group. It was clear that this was an unusual opportunity for Joe to socialise:

They went out to watch the football, have a game of pool and a pint and they brought him back home so that was great, he was that happy, feels as if he’s...it must be great getting away from me because you don’t want to sit with your mother all the time.

There was another contradiction here for mothers who in one respect wanted their child to have all the same growing up experiences as any other teenager but were also grateful that their child was not being exposed to the drink and drugs culture and other high risk behaviour often associated with adolescence. Cameron’s mother perceived that Cameron was missing out by not having a ‘normal’ social life:

In some ways it’s easier because you don’t have to worry about him because he’s not disappearing to the pub or night clubs or whatever but on the other hand that’s what I want for him. So it’s just sort of catch 22, because that’s not a life for him it’s really no life at all.

Lisa’s mother was also very aware of the difference between Lisa’s experience and what might be expected for a young woman of her age:

She's eighteen and a half she should be going out. Usually parents of eighteen and a half year olds are trying to keep them in and Lisa's never out and other friends'll say to me, 'well when you see the boys and girls hanging around the street corner on our street you should think yourself lucky'. So sometimes you think well yes that's fine but is it ever going to change? Will she never be out there with people and meeting people?

Aspirations

In interviews mothers were asked what they would like to see changed. In relation to many issues that were raised mothers talked about what they would like for their son's or daughter's future and also about what the young people themselves aspired to.

In general mothers were realistic in their hopes and expectations. No-one was looking for a 'miracle cure'. All mothers accepted the young person's limitations but also recognised the failings in the support system which hindered their progress. Ryan's mother summed up her aspirations for Ryan as follows:

I want him secure, I want him to be confident and able to handle the minor things that life throws at you and hopefully there will be people round about him that will help him with the major things.

Luke's mother hoped that his siblings would look out for his interests but also wanted to be sure that he would have the support he needed when she was no longer able to care for him:

I have to see that he is going be able to live on his own. I have got to make sure that when we are not here that he has got a place, he's got friends, he is managing this, you know he can feed himself, and all that sort of thing.

Aimee's mother was aware of the difficulties Aimee would face although she knew what she would want for her in an ideal situation:

It would be lovely if there was somebody that loved her and knew how sweet and wonderful she was that would always take care of her. It's hard to think of it and then again, its horrible to think of her

somewhere, where I would be turning in my grave that she was. I don't know how you make that kind of proper provision. I don't know. I mean she will get everything that we've got, but.....

The young people themselves were reported by their mothers as wanting the same things as most other young people: a job, friends, social acceptance, money and a girlfriend or boyfriend. These things were perceived as being more difficult for these young people to achieve but not necessarily as impossible. Logan's mother was confident that Logan would find a partner:

I do believe eventually he will meet somebody, you know, there is somebody out there for everybody.

Daniel's mother was proud of her son, she felt that things had become easier as Daniel had grown up and she was satisfied with the future she saw for him:

He's grown up into such a lovely young man and capable. We can now see Daniel having a future. Daniel's going to have a job. He's going to have a place in the community.

Aidan's mother also saw that Aidan had made progress and more importantly that he was happy:

He seems to be doing okay at college and I am quite happy and he is happy. That is where I see the big difference in him, he is much happier than ever he has been. He goes on to the computer and he chats to all the ones he was at school with, and he has met them in town.

4.7 Summary

The findings from qualitative analysis of the interviews were categorized as representing four main themes: ways of coping; disability identity; support; and hopes and fears for the future. These four themes seemed to capture the reported experiences of the mothers in this study.

Mothers coped in different ways, although for most, acceptance was an important feature. They tried to make sense of the situation and felt that they had learned from

their experiences. They sought to preserve a positive self-image by comparing themselves with others who they considered to be less fortunate. One of the most difficult aspects for mothers of young people with ASD with or without a diagnosis was social acceptance and explaining unusual behaviour to other people.

Most of the young people in this study had mild disabilities and many had suffered the perceived social stigma of being different. Contrasting opinions were expressed by mothers who were grateful for the abilities their son or daughter had but wondered if society might be more accepting if there was a more obvious disability.

Family was the main source of support although family was also often a source of difficulty as some mothers felt family members blamed them for the young person's difficulties and criticized their parenting. Informal supports were also important. Mixed views were expressed about professional support and service provision, with a common sense that adequate information and support was not available.

Mothers were often not sure about how the young person's future would unfold. They had concerns about college and work placements, about long term accommodation and about the young person's social and emotional well-being.

Within the 4 themes the mothers' attempts to help the young person grow into a happy and fulfilled adult, in terms of education, work and personal and social life emerged frequently. In particular, mothers hoped that the young person would become independent or as independent as possible, be accepted by their community, be cared for and supported as necessary and that they would achieve their potential. Although this could be true for any group of parents of young people approaching adulthood, for this group there seemed to be a perception of real barriers to be overcome in achieving these aims and this was a cause of stress and anxiety for many mothers.

In spite of the many difficulties faced by these young people and their mothers, mothers reported that there were also positive aspects to their experiences. Many

mothers described how they had grown in confidence, maturity and become more tolerant through learning to cope with having a child with ASN. Many were proud of their son's or daughter's positive qualities and many had also gained great satisfaction from helping other mothers of young people with ASN. Some felt they had gained a new outlook on life through their experiences.

The themes emerging from this phase of the study and how these relate to research reported in the literature as well as to the quantitative arm of the study will be discussed in Chapter 5.

CHAPTER FIVE - DISCUSSION

This chapter draws together the findings from the quantitative and the qualitative phases of the study. Issues raised are discussed in relation to the main study hypotheses and research questions. Other secondary issues arising through the research are also discussed and the strengths and limitations of the study are described including discussion of the rationale for the combination of qualitative and quantitative methods. Implications for service provision and areas of interest for further research are identified.

5.1 Research Questions

This section will discuss findings from both the qualitative and the quantitative phases of the study in relation to four central research questions. A number of issues have been highlighted throughout this thesis including the initial hypotheses which generated a number of other areas of uncertainty. All of this can be summarised in the following questions:

1. Are there significant differences in family quality of life (FQoL) and maternal QoL between mothers of young people with additional support needs (ASN) and mothers of typically developing young people?
2. Is there a difference in family quality of life (FQoL), maternal QoL and maternal stress between mothers of young people with ASN and autistic spectrum disorder (ASD) and those with ASN but no ASD? If this is the case, then why should this be so?
3. What are the implications for mothers and families of having a young family member with an ASD that has not been diagnosed? Are FQoL, maternal individual QoL or maternal stress affected when there is no diagnosis, either negatively or positively?

4. What are the additional effects on FQoL, maternal QoL and maternal stress of: presence of behaviour difficulties, severity of ASD, young person's age, gender and level of service provision?

5.1.1 Question 1

Are there significant differences in FQoL and maternal QoL between mothers of young people with ASN and mothers of typically developing young people?

Both FQoL and maternal QoL mean scores were found to be significantly higher in mothers of typically developing young people than in each of the 3 groups of mothers of young people with ASN. This remained true across all 4 domains (physical health, psychological well-being, social relations and environment) of the measure of maternal QoL, the Whoqol-bref. Having a son or daughter with social communication difficulties with or without a diagnosis of ASD had an additional negative effect on scores for mothers of young people with ASN.

These findings are consistent with the published literature in which mothers of young people with intellectual and developmental disabilities suffer higher levels of stress than mothers of typically developing young people and also that showing that mothers of young people with ASD suffer higher stress levels than mothers of those with other types of disability (Wolf *et al* 1989; Stores *et al* 1998; Fombonne *et al* 2001; Nachshen and Minnes 2005). Families of young people with disabilities have been reported to be more likely to be socially disadvantaged and this has been cited as a possible contributor to the lower QoL and higher maternal stress reported for this group (Emerson 2003a). However, in this study there were no significant differences in SES amongst the four groups suggesting that in this case SES was not affecting group differences. In addition, many of the published studies refer to groups of children with moderate to severe learning disabilities including those with physical disabilities and significant health problems. In contrast, the young people in this study had difficulties or disabilities that were relatively mild, some had IQ scores within the normal range and none had significant physical disabilities. Yet in spite of this, the mothers of these young people still reported a significantly lower quality of

life for themselves and for their families than the control group of mothers of typically developing young people.

The difference in FQoL between families of young people with ASN and families of typically developing young people was sustained when FQoL was considered from the perspective of fathers and young people themselves, but not when considered from the perspective of siblings. This may have been because the numbers within each group of other family members were too small to give meaningful results, or it could have been because the fathers and young people were more strongly influenced by the mother when completing the questionnaire. It is also possible that the siblings of this group of generally mildly disabled young people genuinely did not suffer a significant degree of negative impact on their quality of life.

This interpretation was supported by the findings from the qualitative interviews. Although mothers reported that siblings would be expected to take some responsibility for their brother or sister in the future, this was not described as being something that the siblings resented or resisted. Siblings, especially as they grew older, were reported as being supportive of their brother or sister with ASN. Only one mother reported a difficult stage as a younger sibling had to come to terms with the fact that his older brother had ASN. This child had felt angry that his brother had not lived up to his expectations by being unable to take on the role of protector; the mother also reported that the younger brother had felt ashamed of having a brother who was different. Published research on the adjustment of siblings of young people with disabilities has reported conflicting results with positive as well as negative outcomes described (Fisman *et al* 2000; Hastings *et al* 2005; Giallo and Gavidia-Payne 2006).

5.1.2 Question 2

Is there a difference in FQoL, maternal QoL and maternal stress between mothers of young people with ASN and ASD and those with ASN but no ASD? If this is the case, then why should this be so?

The results from the quantitative phase of the study suggest that there are differences in FQoL between the two groups, with mothers of young people scoring above the ASD screening cut-off (groups 2 and 3) reporting lower FQoL than those with ASN scoring below the cut-off (group 1). Higher levels of challenging behaviour in groups 2 and 3 may be a contributing factor but partial correlations showed that statistically this does not account for the difference. Higher levels of maternal stress and lower maternal individual QoL are also both associated with lower FQoL and with higher SCQ score, i.e. possible presence of ASD, but the direction of causation for both these variables is likely to be complex to determine.

The qualitative phase of the study found that mothers of young people with positive SCQ scores (with or without an ASD diagnosis) were more likely to report difficulties in explaining the young person's additional needs to others than mothers of young people with non-ASD ASN. Findings from the interviews suggested that the young person's unusual behaviour and social communication difficulties may be an additional stressor for mothers and young people for two reasons. Firstly, the young people in this study generally had mild disabilities with few outward signs of their difficulties and as a consequence, people they came into contact with might not appreciate their limitations and therefore be less likely to be tolerant of unusual behaviour. Secondly, those with social communication difficulties are more likely to struggle with understanding the rules of social behaviour and to behave in a way that is perceived as, at best, socially odd and possibly as unacceptable. In such circumstances behaviour could be interpreted as rudeness or bad manners. This experience was reported by mothers of both groups of young people with positive ASD screening scores although not for the group with non-ASD ASN. Related to this experience, mothers of the group with positive ASD screening scores also reported feeling inadequate or blamed for the young person's behaviour, with the perceived implication that they were not a 'good mother'. For these mothers who had described struggling to do the very best for their child, this was reported as being particularly hurtful. Mothers of young people with non-ASD ASN again did not report such experiences and it seems likely that these young people with mild disabilities would be more likely to behave in a socially acceptable way and more

able to take part in community life without attracting unwanted attention. These reported perceptions may be contributing factors to the lower FQoL scores for mothers of young people with positive ASD screening scores, as well as the reported higher levels of stress and lower levels of maternal QoL for this group in comparison to mothers of young people with non-ASD ASN.

5.1.3 Question 3

What are the implications for mothers of having a young family member with an ASD that has not been diagnosed? Are FQoL, maternal QoL or maternal stress affected when there is no diagnosis, either negatively or positively?

It had been hypothesised that mothers of young people with positive ASD screening scores but without a diagnosis of ASD would be additionally disadvantaged compared with those who had received a diagnosis and as such would be benefiting from more appropriate services and support. However, this was not supported by the results of the quantitative phase of the study which showed no statistical differences between the two groups scoring above the ASD cut-off in terms of individual or family QoL or of maternal stress, whether with or without a diagnosis of ASD.

Possible reasons for the continuing increased stress for mothers of young people with a diagnosis of ASD were explored in the qualitative phase of the study. In spite of having a diagnosis, these mothers reported receiving inadequate services and support and facing a lack of understanding from others. The process of receiving a diagnosis for the young person was often described as a protracted struggle, with much disagreement amongst professionals and often, at the end, an unsatisfactorily vague diagnosis. This finding is consistent with the published literature, see e.g. (Brogan and Knussen 2003).

Mothers in the qualitative study reported that information about the diagnosis was not passed from one professional group to another (for example from primary to secondary school) and that both professionals and family members did not always accept the validity of the diagnosis. In general there was a perception of lack of understanding and acceptance by others and even when the young people had

received a diagnosis, appropriate help was not always available. It is possible that these reported experiences may explain why the mothers who had received a diagnosis did not necessarily experience lower levels of stress or higher levels of QoL as a consequence.

However, mothers reported positive outcomes from having a diagnosis and also described positive relationships with individual professionals who had been particularly supportive to their family. Mothers also reported being relieved to receive the diagnosis and in some cases this had gained them access to support organisations and to other parents of young people with ASD. This was generally felt to be useful.

As there were no statistically significant differences in the main study variables between the groups scoring above the ASD cut-off with and without a diagnosis, the question remained as to the reasons why certain young people had received a diagnosis while others had not.

It had been expected that those with more severe symptoms of ASD would be more likely to receive a diagnosis, but this was not found to be the case. The 2 groups scoring above the ASD cut-off did not differ significantly in terms of severity of ASD (SCQ score).

The groups also did not differ significantly in terms of intelligence (IQ score) although again it had been expected that those young people with ASD and higher IQ might have been more able to compensate for their difficulties and this in turn might have lead to some avoiding being given a diagnosis. Again, this was shown not to be the case, however.

Another possible reason for differences in rates of diagnosis could have been socio-economic status with the more affluent and educated parents being more able to access services, again in this study this was shown not to be the case as the four groups did not differ significantly in terms of SES.

Challenging behaviour (CBCL score) was associated with higher SCQ scores and an early hypothesis had been that those young people who had received a diagnosis would be likely to show higher levels of challenging behaviour, as this might increase the likelihood of the young person coming to the attention of a clinician and thus undergoing assessment. However, again this pattern was not found in this sample, with no significant differences in CBCL score between the 2 groups. However, mean scores for the 8 subscales of the CBCL were also examined for group differences and although there were still no significant differences between the 2 groups, the pattern of scoring suggested that there might have been a difference between the groups with and without a diagnosis in the subscale 'thought problems'. This subscale, which includes behaviours that are related to schizotypal characteristics, showed a different scoring pattern to the others in that this was the only subscale in which the group with an ASD diagnosis (group 2) scored higher than the group without an ASD diagnosis (group 3). In the other 7 subscales the two groups scores were either almost the same or group 3 scored slightly higher (see Fig. 3.11, p.77).

This finding suggested that group 2 may be different from group 3 in respect of schizotypal characteristics with the possibility that these are behaviours which increase the likelihood of young people being referred to clinicians who may then make the diagnosis of ASD.

Although assessment and diagnostic measures used today are more focused on behaviour and developmental assessment rather than measures of psychopathology, it is possible that professionals may still be influenced by previous theories which tended to relate autistic features to psychiatric disorders. For example, an early definition of ASD in childhood was childhood schizophrenia (Rutter 1978).

The only striking difference between the 2 groups with and without a diagnosis was the gender ratio. Of all those scoring above the ASD cut-off, 83% were male; this is close to the reported ratio of 4:1 males to females (Ehlers and Gillberg 1993).

However, in the group with a diagnosis, 17 of the 18 young people (95%) were male. This suggests the possibility that males are more likely to receive a diagnosis. This could be because clinicians expect that an individual with ASD is more likely to be male and so tend to discount symptoms in females, or it could be because of differences in the behavioural phenotype of ASD in males and females, with greater recognition of male ASD-related behaviours. The patterns of scoring on the CBCL and its subscales were also examined to look for gender differences in behavioural profiles which might contribute to the likelihood of a young person with ASD receiving a diagnosis. It was found that the total scores did not differ significantly between males and females, nor did any of the subscales except for the one measuring delinquent behaviour where male scores were significantly higher. In most subscales male scores were slightly higher and this included the subscale 'thought problems' which had shown the difference between the 2 groups with and without an ASD diagnosis. These findings do not provide any substantive evidence to support the hypothesis that males are more likely than females to receive a diagnosis of ASD because of differences in their behavioural profile. However, it may still be the case that clinicians have different expectations of male and female patients.

It is also possible that young people are more or less likely to receive a diagnosis based on local practices in different geographical locations. In this study participants were recruited from across Scotland, representing many different health and education authorities and urban as well as rural areas. The numbers drawn from each location were too small to draw conclusions, however. While in most areas there was a fairly even divide between those with and without a diagnosis, in one area, Greater Glasgow, there was a considerably greater proportion of young people scoring above the ASD cut-off who did not have a diagnosis. Further investigation involving much larger population samples would be required before concluding that differences were due to regional variations on diagnostic procedures. However, Knussen & Brogan (2002) did find variations in their study of professionals' practice in the disclosure of diagnosis of ASD in 3 areas of Scotland which suggests the possibility that other differences in diagnostic profiles may also be present.

Another difference amongst the geographical locations included in the study related to the numbers of young people in each group attending special or mainstream school. Again the numbers were small and so results can only identify areas of interest for future investigation. It is known that urban areas, particularly the cities of Edinburgh and Glasgow, have a greater degree of special school provision than other areas of Scotland. In many rural areas there is little alternative to the local school unless the child requires specialist provision, with this often outside of their home area, for example in a residential school.

Initially it had been expected that those attending special school would be more likely to be those with a diagnosis and that those within mainstream education might include young people who were not receiving special education because the more specific nature of their difficulties had not been identified. However, the range of educational provision and the current policy of inclusion whereby young people are now all expected to attend mainstream schools, along with the degree of special provision now available within the educational mainstream makes these comparisons difficult to interpret. In this sample, of those still attending school, group 1 (non-ASD ASN) had the lowest proportion attending special school (46%), group 2 (ASD positive with ASD diagnosis) had an increased proportion (65%), while group 3 (ASD positive but without diagnosis) had the highest proportion, with 79% of this group attending special school. This pattern showed the same trend as the levels of challenging behaviour and it is likely that this may be one of the factors affecting the decision regarding placement at mainstream or special school.

It is also possible that the groups with and without diagnosis did not differ in scores on the main study variables because there is such wide variation in the circumstances and experiences of families, with much overlap between the 2 groups. In some cases young people received a diagnosis of ASD after initially being recruited to the larger main parent study without a diagnosis. Levels of diagnosis also varied from, a parent reporting having been given a vague diagnosis such as 'autistic tendencies' to those receiving a more definite diagnosis after a reliable assessment process.

There were also 2 cases where mothers reported that the young person had been given a diagnosis of ASD although they scored below the ASD screening cut-off for this study. In each of these cases the diagnosis, both of Asperger's syndrome (AS), had been given after recruitment to the parent study and there was some reported ambiguity about the diagnosis.

These examples demonstrate how the groups with and without a diagnosis tended to have similar experiences, and that the level of information and support available after diagnosis was also very variable and thus likely to affect QoL and maternal stress. However in spite of the lack of significant differences in scores on the main study measures, those interviewed did report positive perceptions of the benefits of having a diagnosis, with this almost universally welcomed. Mothers with and without a diagnosis talked about the difficulties of securing appropriate services when the young person did not have a label to describe their difficulties. This was an issue in particular for those moving from child to adult services and seeking to negotiate the benefits system including eligibility for various supports related to education, work, housing, social support or independent living allowance.

5.1.4 Question 4

What are the additional effects on FQoL, maternal QoL and maternal stress of: severity of ASD, presence of behaviour difficulties young person's age, gender and level of service provision?

There was a significant positive correlation between SCQ score (level of ASD) and maternal stress which remained significant, although reduced, when controlling for scores of challenging behaviour. The reduction in significance suggested that levels of challenging behaviour were also having a negative effect on maternal stress, although the effect was not strong enough to account for group differences. CBCL (challenging behaviour) score was also shown to be significantly higher for those scoring above the cut-off for ASD, although there was no significant difference between the groups with and without a diagnosis. This suggests that higher levels of

challenging behaviour are not the factor which makes receiving a diagnosis more likely for young people with ASD.

In this study there were no effects of age. Age did not correlate with any of the other study variables. Nor did gender correlate with any variable other than ASD score, with more males than females represented in the groups with higher SCQ scores. This could be expected as ASD is more common in males and Asperger's Syndrome (AS), the diagnosis of 6 of the young people, affects an even higher ratio of males to females. However, gender did not correlate with stress, or family or individual maternal QoL scores, although previous research has suggested that parenting male children is more stressful than parenting females (Emerson 2003a; Bromley *et al* 2004).

The measures in this study suggested that number of service contacts did not differ significantly amongst the 3 groups of mothers of young people with ASN and that although service usefulness was reported as being lower in group 3, this difference was not significant. In interviews mothers often described their difficulties in getting the levels of support required for their son or daughter and in questionnaire replies 25% of mothers reported having no contact with services during the past month. However, there were not obvious differences in the reported perceptions of families with little or no service contact compared with those with high levels of service contact. There was also no correlation between the number of contacts with services or the perceived usefulness of service provision with FQoL or maternal QoL.

There was however a significant positive association between the number of service contacts and maternal stress. This of course does not tell us anything about the direction of causation, only that as the number of reported contacts with services increased, maternal stress also increased. Levels of support have more typically been reported in the literature as a mediating factor in levels of stress in mothers of young people with disabilities (Bromley *et al* 2004; Hassall *et al* 2005). The higher number of contacts reported by mothers in this study however could be a reflection of their increased needs and therefore help-seeking behaviour, that is, the association could

be an indicator of the increased level of support needed by the group with higher stress levels rather than a measure of the effectiveness of the services provided; the lack of any significant correlation between service usefulness and maternal stress scores would support such an interpretation. Alternatively, it is possible that the presence of a large number of professional services is in itself a stressful experience for mothers.

It also has to be acknowledged that the very simple measure used to assess service contact and usefulness may not have been sensitive enough to detect any differences across groups.

5.2 Secondary Issues

5.2.1 Mothers' perceptions of stressful issues

The issue most commonly chosen as the most stressful for mothers was 'creating and/or finding opportunities for the young person to make friends and participate in activities'. This suggests that social acceptance was an important issue for all 3 groups. This could be related to the age of the young people who were all approaching adulthood. This is often reported as a difficult developmental stage but may be particularly so for this group of young people with some additional support needs but whose needs are relatively mild, leaving them aware of the differences between themselves and their typically developing siblings or peers. The next most stressful issues were also related to this transitional stage, for example, finding work or employment and planning for the future, particularly concerns relating to accommodation. In both these areas mothers reported a degree of frustration in relation to perceptions of lack of provision, poor access to information and poor communication with and amongst service providers, with all these leading to considerable concern over possible future outcomes.

5.2.2 Unmet needs

In the qualitative interviews mothers were specifically asked what they would like to see changed to make things easier for themselves or others in the future. in relation to

each of the chosen stressful issues. Commonly reported unmet needs were: lack of support, lack of access to information and poor co-ordination of services. Mothers felt that did not know where to get the information they needed and they did not know where to turn for help or advice in a crisis; they also felt that the different services they dealt with were poorly co-ordinated, and did not communicate well with either families or with each other, leaving it difficult to plan for the future.

Mothers' suggestions for improvements in services and support included: increased awareness and understanding of ASN and ASD from professionals, family and the public, possibly through education and public awareness programmes. Mothers also highlighted their perceptions of the need for increases in financial support for services to improve staffing and facilities, such as greater availability of respite services and increased numbers and availability of social workers, a professional grouping reported to be particularly difficult to contact due to understaffing. Wider changes in the attitudes of society and in the government's approach to meeting the needs of young people with ASN and their families were also often suggested. These findings were compatible with reports from the social research literature on outcomes for families of children and young people with a range of disabilities which highlight unmet needs for respite care, social and financial support and flexible, well co-ordinated service provision (Joseph Rowntree Foundation 1999; Sloper and Beresford 2006; Beresford *et al* 2007).

5.2.3 Maternal characteristics

Another secondary issues to emerge was the possible role of maternal characteristics in explaining the differences in perceptions of stress or in reactions to stressful situations and the question of whether maternal characteristics may have an influence on why some young people scoring above the ASD cut-off received a diagnosis and others did not.

Mothers of children and young people with ASD have been reported to be at increased risk of stress or depression due to the effect of the broader autistic

phenotype as parents are thought to exhibit certain traits related to the condition even although they do not themselves meet diagnostic criteria (Duarte *et al* 2003). This could be a further explanation for the increased level of difficulties reported by mothers of young people in this study as well as their higher stress and lower QoL scores than mothers of young people with non-ASD ASN.

Other factors affecting outcomes for mothers and influenced by maternal characteristics could be the coping strategies they employ. Coping was one of the major themes emerging from the interview data and included a wide variety of descriptions of different coping strategies that mothers had successfully and unsuccessfully used. Previous research has investigated coping methods and their relation to levels of stress (Gray 2006), with much of this suggesting that coping strategies based on acceptance and positive reappraisal are associated with lower stress levels than strategies centred on escape/avoidance (Hassall *et al* 2005; Hastings *et al* 2005). Mothers in this study reported using all of these strategies. Many quotes represented acceptance, such as ‘just getting on with it’, and positive reappraisal, such as ‘things could be worse’.

Positive perceptions as well as being recognised as a protective mechanism have also been reported as an under-researched area (Hastings 2003). In this study many positive perceptions were expressed, such as the personal gains in terms of maturity, tolerance and understanding, strength of character as well as the opportunity to give something back by helping others. Also, many parents were proud of the young people and what they had achieved in spite of their difficulties. The young people’s characters and family interactions were also often highly valued by parents and family members. Although the numbers taking part in interview were small and it was not possible to measure this statistically, the mothers who expressed positive perceptions about their son or daughter tended to have lower stress scores than those who did not.

5.2.4 Approach to provision of services and support

The ability to accept what you cannot change is a familiar concept in our society, and the lowering of stress levels as mothers come to acceptance of their situation is understandable. However in this study even the mothers with low stress scores, who accepted their child's difficulties and had positive perceptions about them and their abilities, identified unmet needs. The final question in the semi-structured interview relating to what mothers would like to see changed and that would make things easier generated as many responses from mothers suffering lower stress as from those suffering higher stress, suggesting that practical initiatives are required as well as supportive measures for those showing the greatest stress.

Although the young people in this study had mild disabilities, for most of them these were difficulties that they would have to learn to live with; they would be able to improve their abilities and understanding but they would always be likely to need some extra support in their day-to-day lives. The mothers in this study were fully aware of this and none reported seeking any treatment or intervention that it was thought might change the young people's fundamental difficulties. This represented a high degree of acceptance and while measures to improve coping and acceptance may be useful in the management and support of families, changes in the way that services are provided for this group may be of more lasting benefit and in themselves may help to alleviate stress.

While improving coping strategies represents an internal solution to the problem, adapting the environment represents an external solution. The internal solution implies that the source of the difficulties lies within the individual, while the external solution, with its focus on adapting the environment, implies that the source of difficulties is within the physical or social environment the individual inhabits. The latter view reflects the perspective of the disability movement which views disability as a social construct and suggests that outcomes for those with disabilities, whatever the kind, would be different if physical, societal and attitudinal barriers were removed (Shakespeare 2000). Turnbull et al (2004) describe an American view of the same concept as the 'New Paradigm of Disability' which focuses on fixing the

system in contrast to the 'Old Paradigm' which emphasised fixing the individual or the family.

In the context of this study population neither approach on its own will entirely resolve their difficulties. However, aspects of each could have a useful contribution to improving QoL for these mothers and young people and their families. The notion of more effective coping strategies could be useful for mothers; for example with examples of effective techniques passed from one mother to another through support groups or telephone help lines, in this way, a successfully coping mother could help one who was struggling to reappraise her situation and find a more positive perspective. Encouraging a mother to adopt or recognise a more positive perspective in her own situation could perhaps enhance coping and reduce stress. The practical barriers faced by these young people and their families are also potentially amenable to change, however, and although the Scottish Executive has already made recommendations addressing these issues in a way that theoretically should be helpful (Scottish Executive 2000a), the roll out of these policies seems to have been slow. Addressing these difficulties through fast-tracking the introduction of Local Area Co-ordinators (LAC) would seem to be a very necessary step at this stage and one that could be undertaken without great expenditure or renegotiation of priorities as the basic underlying approach has already been approved, has been implemented to a degree and research evaluating the effectiveness of the policy has been undertaken (Curtice 2006; Stalker *et al* 2007).

None of the mothers interviewed in this study mentioned local area co-ordination although some did live in areas where the service was theoretically available. In some cases a professional had helped to liaise with other services and had made enquiries about services that the young person might need, but when this occurred it seemed to be perceived as the actions of a particularly supportive individual who had gone beyond what would normally be expected, rather than as someone fulfilling a specific professional role. It was not clear whether any of the professionals identified were employed as local area co-ordinators.

5.3 Design of the study

There is sometimes a perceived divide between quantitative and qualitative research methods which to some extent reflects the contradictory underpinning value systems of each approach. Quantitative methods are generally based on a positivist world view which perceives the nature of reality consisting of truths which in principle can be known to the inquirer. Qualitative approaches tend to take an inductive approach, seeing truth as a more subjective concept accessible only through an individual's perception of reality. Although there has been some debate amongst researchers about the compatibility of the two approaches, in this study a pragmatic stance has been adopted which recognises that each approach is appropriate for different sorts of questions and produces different sorts of answers (Bryman 1984; Lincoln and Guba 1985; Polit and Hungler 1999). The combining of methods in this study was driven by the needs of the research questions rather than by methodological ideology.

Barbour (1999) describes the following reasons for combining quantitative and qualitative approaches: to explore the mechanisms by which variables are connected; to provide a context for quantitative findings; to bring more than one source of data to the understanding of concept; and to qualify findings. Qualitative methods are also recognised as a useful way of generating directions for future research.

Quantitative methods are used to provide measurable evidence and meaningful comparisons of outcomes for groups in a study in a way that may be generalisable to a wider population. However, while the questionnaires used in this study elicited responses to pre-selected items which were quantified according to pre-determined criteria, the interviews allowed the respondent the freedom to express their opinion about issues that were important to them and to describe their attitudes or feelings. For example, when questionnaire responses indicated that a particular issue was extremely stressful for the respondent, the interview helped to provide greater insights into how the mother perceived the issue and an explanation of why it was stressful for her. The inclusion of qualitative interviews therefore allowed a richer description of the experiences identified as stressful by the questionnaire responses. Analysis of these data aimed to complement the analysis of quantitative findings,

allowing for a more complete exploration of the issues being investigated. One benefit of using the Family Stress and Coping interview (FSCI) as a measure was that it contained both quantitative and qualitative sections.

While the quantitative phase can identify statistically significant differences amongst the study groups, the qualitative phase can provide insight into the life experiences and perceptions of the mothers in this study. By combining methods, this study aimed to achieve a fuller picture of the life experiences for mothers of young people with ASN.

It is recognised that the nature of qualitative research is that it seeks to represent an 'emic' or 'insider' perspective (Polit and Hungler 1999) i.e. the point of view of the person being interviewed rather than the researcher. The limitation is recognised that this perspective may be unclear or ambiguous and may be affected by the participant's interaction with the researcher or by the image the participant wishes to portray. Through the interview the researcher has access to one particular interpretation of the situation being experienced and this is recognised as a feature of qualitative research: 'truth' is not seen in the absolute sense that is implicit in positivist approach used in most quantitative research, but rather truth is considered as something that can be interpreted differently by different people and in different circumstances (McCann and Clark 2003). However, the inclusion of quantitative data can provide evidence to support the generalisability of qualitative findings.

5.4 Limitations and Strengths

5.4.1 Limitations

The limitations of this study include the fairly small sample size and in particular the small numbers of other family members providing data, making statistical analysis of data from other family members difficult. Also, there was a representation of many areas of Scotland and while this was in itself a strength, it also necessarily reduced the numbers from each area, making it difficult to draw conclusions about any

differences which might stem more from differences in local practices rather than from individual differences.

Another difficulty was the broad range of ability of the young people recruited. Again this was as a result of the sample being a population sample recruited through education services, where IQs are not measured and perception of the concept of learning difficulty, learning disability and additional support needs are used at times interchangeably and often differently from the use of these terms in clinical settings. These factors led to the recruitment of a fairly heterogeneous sample, particularly in the group of young people with diagnosed ASD, where there was a broad range of IQ scores. However, as the young people were recruited into the main parent study on the basis of teacher report and as their IQ was not measured until after recruitment, the sample has been defined by having been identified by educational services as having additional support needs, rather than as having an IQ within a certain range. This necessarily means that some of the young people did not meet the criteria for a diagnosis of learning disability.

5.4.2. Strengths

A major strength of this study is that the sample was recruited through the education system rather than being clinically derived, making this sample much more likely to represent the general population of people with ASN.

The study also benefits from having a population drawn from many different areas of Scotland ensuring a broad representation and avoiding the possibility that findings are due to unusual local circumstances.

Having a control group of age-matched typically developing young people allowed a useful comparison with mothers of young people without ASN. The inclusion of data from other family members, including siblings, served in one respect to ensure that family perspectives were considered, recognising that the mother's opinion might not be shared by other family members and also adding weight to the mother's opinion when family members agreed.

Another positive aspect of the study design was the inclusion of questions in the semi-structured interview which focused on the mother's strengths, recognising her expertise and valuing her opinions by asking about her perceptions of available services and what she would like to see changed. These questions in the qualitative section of the FSCI were explicitly included with the aim of being empowering for the mother (Nachshen 2003). Such questions were valued by mothers who often reported feeling that they had little opportunity to voice their opinions or that their views were not being taken into account by the professionals with whom they had been involved.

Another strength of this study is the use of both quantitative and qualitative methods. Each approach provides a different perspective on the issues being explored. The findings from each phase support each other, and taken together, provide a more meaningful account of the participants' perceptions and experiences.

5.5 Implications

5.5.1 Service provision

There is a clear need for improved service provision for young people with ASN and their families. The more widespread implementation of local area co-ordination services which local authorities are being encouraged by government to implement, could be a key provision in meeting the perceived needs of the mothers and families in the study, addressing many of the main issues raised. Such services could also enhance aspects of coping if the LAC was able to provide support by for example, introducing new coping strategies or by linking the mother with a support group.

Most of the mothers expressed positive attitudes to their sons and daughters but were frustrated at the perceived barriers to their progress. Mothers' suggestions for improvements did not seem unrealistic nor even likely to be difficult to achieve in practice. Mothers' suggestions included ideas in line with government

recommendations, based on previous research on family needs and wishes, which are currently being implemented across Scotland. The findings from this research support these recommendations from the 'The Same as You?' Review (Scottish Executive 2000a), in particular the introduction of local area co-ordination but also demonstrate weaknesses in their implementation. Reasons for this may include wide variations across the country both in the interpretation of recommendations as well as in implementation procedures (Short life working group on local area co-ordination 2002).

A recent evaluation of LAC implementation (Stalker *et al* 2007) has reported positive outcomes both from the perspective of local area coordinators themselves and also from the individuals and families that they support. Areas of achievement identified were an improvement in overall QoL and also in some specific areas including transitions to adulthood. These findings add further evidence to the conclusion that local area coordination would be of great benefit to the young people and families whose experiences and perceptions are reported in this thesis. The fact that so many mothers reported unmet needs and suggested solutions that could be met by provision of an LAC suggests that this is the right policy but that it is not yet being effectively implemented in all areas of Scotland. Stalker *et al* (2007) conclude their evaluation of local area coordination with recommendations for an increase in the number of LAC posts with provision of appropriate training as well as updated and improved practical guidelines for implementation to ensure that all areas of Scotland have adequate and equal access to this service .

A reason why some of the families studied here were not being supported by a LAC although they lived in areas where this service was theoretically available, may have been that the young people were not a clinical sample but were recruited through the education system and thus may not have been known to the service providers who could direct them to the LAC. The young people with ASN who participated in this study may also not necessarily meet the criteria for intellectual disability (IQ<70) and as LAC is a service for people with learning disability and their families they may not be eligible for this service. As suggested by the findings in this thesis, this

may underlie the reported high levels of maternal stress and lower family QoL for this group of mothers of young people with ASN. The relatively mild levels of disability and at times poor understanding of underlying difficulties may also account for some of the other stressors reported, such as: inadequate educational provision, poor social acceptance, and other problems in developing self-image and self-esteem.

5.5.2 Further research

The findings from this study suggested many areas for further research. One main issue would be further investigation of the implementation of LAC services in order to determine why this service may not be reaching all those who might benefit from it and to identify ways of improving professional practice in this area, although some of these issues have been addressed in the recently published evaluation (Stalker *et al* 2007).

Another interesting related area for investigation would be a study of similar design to that reported in this thesis, examining the same issues (maternal stress, family QoL, maternal QoL and satisfaction with service provision) for a sample of families who were being supported by a local area coordinator.

A larger study looking at possible variations across different geographical locations in approaches to diagnosing ASD and to provision of educational support and at how these relate to service-user satisfaction would give further insight into this area. Also, links between service contacts and their usefulness and family QoL and maternal stress could be further investigated with a larger sample.

The perspectives of other family members including young people with ASN would also benefit from further exploration in a larger sample group. A longitudinal study exploring outcomes as young people with ASN grow up and looking at perceptions of family members and satisfaction with service provision would be of particular help in increasing understanding of the experiences of such families in this important transitional period.

5.6 Conclusions

This study clearly showed that in spite of the relatively mild nature of the difficulties faced by the group of young people with ASN whose families participated, the effect on family QOL and maternal stress was significant. The additional stress which results from the young person having social communication difficulties was also very apparent.

Despite mothers reporting that they valued having a diagnosis for the young person, the results of this study suggest that having a diagnosis of ASD does not have a significant effect on FQoL, maternal QoL or maternal stress for mothers of young people scoring in the ASD range. Suggested reasons for this are that the diagnosis was not always satisfactory and did not necessarily gain the families access to the support they needed. Mothers also perceived difficulties with social acceptance for the young people, whether or not they had a diagnosis, often reporting that others did not understand or accept the implications of a diagnosis of ASD and were intolerant of the young person's difficulties.

The mothers of all 3 groups of young people with ASN expressed a level of dissatisfaction with available services and supports and made several suggestions for service improvements. Mothers' perceptions and expectations were not unrealistic, in general they were very accepting of the young peoples' limitations. Many of the unmet needs identified would not be hard for service providers to meet and in fact are those already identified by policy makers as central to effective service provision.

Mothers of young people with ASN in this study wanted the same for their sons and daughters as most mothers, for them: to grow up to be happy and secure, to find something they wanted to do that gave them satisfaction, and to have people around them who cared about them and were able to support them. However, these mothers perceived many barriers to the achievement of these aims, chiefly a lack of appropriate support, specifically defined in terms of well co-ordinated services, and a lack of understanding and social acceptance, not only from their own communities but also often from friends and family.

There are clearly no perfect answers to the difficulties facing the mothers in this study. Obviously many would benefit from the development of a more accurate definition of the range of behaviours and characteristics within the autistic spectrum and from more reliable and consistent methods of diagnosis. However the key issue highlighted in this work and the one which would seem to have the greatest potential to provide positive benefits for these young people and their families, is the improved implementation of local area co-ordination services with the potential to provide much-needed support for such families.

Appendix 1

Abbreviations

Abbreviation	Meaning	Definition
ADHD	Attention Deficit Hyperactivity Disorder	Condition characterised by inattention, lack of concentration, impulsivity and hyperactivity
ANOVA	Analysis of variance	Statistical technique used to compare several means
ASN	Additional Support Needs	Term used in the Scottish education system referring to young people with educational difficulties for a variety of reasons
AS	Asperger syndrome	Condition within the autistic spectrum often without ID or language delay
ASD	Autistic Spectrum Disorder	Range of disorders characterised by the triad of impairments
CBCL	Childhood Behaviour Checklist	Measure of challenging behaviour (Achenbach 1991)
CP	Cerebral Palsy	Group of conditions characterised by movement problems caused by brain damage or abnormal development often before or during birth, often associated with ID
DAMP	Deficit in Attention, Motor control and Perception	Condition based on concomitant ADHD and developmental co-ordination disorder in children (Gillberg 2003)
FE	Further Education	Post school education at less than degree level
FQoLS	Family Quality of Life Survey	Measure of family quality of life (Park 2003)
FSCI	Family Stress and Coping Interview	Measure of maternal stress (Nachshen 2003)
ID	Intellectual Disability	Also described as learning disability (UK) or mental retardation (US), usually defined by IQ<70
IQ	Intelligence Quotient	Score on standardised measure of general intelligence
LAC	Local Area Co-ordination	Service approach recommended by the Scottish Executive to support people with ID and their families
MMR	Measles Mumps Rubella	Combined measles, mumps and rubella vaccine
MRI	Magnetic Resonance Imaging	Non-invasive technique producing images of the inside of an object, used to scan the human brain

Abbreviation	Meaning	Definition
MRC	Medical Research Council	Publicly funded UK organisation supporting research into human health
NAS	National Autistic Society	Charitable organisation promoting the rights and interests of people with ASD
OCD	Obsessive- Compulsive Disorder	Anxiety disorder characterised by intrusive thoughts, images or worries and/or repetitive non-functional behaviours or acts
PDD	Pervasive Developmental Disorder	Group of conditions including autism, Asperger syndrome, childhood disintegrative disorder, Rett's syndrome and atypical autism or PDD-NOS
PDD-NOS	Pervasive Developmental Disorder- Not Otherwise Specified	Also known as atypical autism, often with later onset and not meeting criteria for full clinical presentation of autism
PHIS	Public Health Institute of Scotland	National agency for improving Scotland's health
PiP	Partnership in Practice	Recommendation of the Scottish Executive for co-ordinated service provision
QoL	Quality of Life	Individual's perceptions of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations, standards and concerns (WHOQOL Group 1998)
SAY	The Same as You?	Scottish Executive review of services for people with learning disabilities (Scottish Executive 2000a)
SCLD	Scottish Consortium on Learning Disability	Organisation set up on recommendation of Scottish Executive to oversee implementation of SAY recommendations
SCQ	Social Communication Questionnaire	ASD screening questionnaire (Berument 1999)
SIRQL	Special Interest Group- Quality of Life	Subgroup of International association for the scientific study of intellectual disabilities
St Dev	Standard Deviation	Statistical measure of how well the mean represents the data set
SWB	Subjective well-being	An individual's sense of their personal well-being
WHO	World Health Organisation	United Nations specialised agency for health

Appendix 2

Measures

1. Parent questionnaire

Section 1 contains the measure of service contacts and usefulness

Section 2 contains the family Stress and coping interview (FSCI)

Section 3 contains the measure of maternal QoL, the Whoqol-bref

Mothers of the control group only received Section 3, 'Your Quality of Life' (the Whoqol-bref)

2. Family quality of life survey (FQoLS)

Questions 22-25, relating to disability issues were removed for families of the control group

3. Childhood behaviour checklist (CBCL)

Data collected for the larger parent study

4. Social communication questionnaire (SCQ)

Data collected for the larger parent study

1. parent questionnaire

Quality of Life in Families of Young People with Additional Learning Needs

Thank you for agreeing to complete this questionnaire.

The following three sections ask about:

1. The amount of services your son or daughter receives and how useful these services are
2. Issues which cause you stress
3. Your feelings about your quality of life

It is important that you answer as many questions as you can, but please feel free to skip any questions that make you feel uncomfortable. If you are unsure about which response to give to a question, please choose the one that appears most appropriate. This can often be your first response.

Section 1: Level of service provision

In the past month, how many contacts have you had with each of the following services related to the care of your son or daughter who is helping with this study? A contact could be a face-to-face meeting, a telephone conversation or a letter from the service provider. Please write the number of contacts in the first column, then circle 0, 1 or 2 to indicate how useful the contact has been.

Service	How many contacts?	Not at all useful	A little useful	Extremely useful	Comment
Education		0	1	2	
Social Work		0	1	2	
Family Doctor (GP)		0	1	2	
Community Nurse		0	1	2	
Other therapist (please state which)		0	1	2	
Voluntary service		0	1	2	
Other		0	1	2	
		0	1	2	

Section 2: Stressful Issues

How stressful are the following issues for you today?

Please circle the number that best describes how stressful each issue is for you.

	Issue	Not Stressful	Mildly Stressful	Moderately Stressful	Extremely Stressful
1	The diagnosis of _____ as having additional learning needs	0	1	2	3
2	Explaining to others about _____'s additional learning needs	0	1	2	3
3	Your feelings about the cause of _____'s additional learning needs	0	1	2	3
4	Dealing with friends/family/people in the neighbourhood on a day-to-day basis	0	1	2	3
5	Dealing with doctors and other allied health professionals	0	1	2	3
6	Dealing with legal professionals	0	1	2	3
7	Dealing with _____'s teachers and the educational system	0	1	2	3
8	Creating and/or finding opportunities for _____ to make friends and participate in activities	0	1	2	3
9	Deciding on the best level of integration for _____	0	1	2	3
10	Making the decision concerning accommodation in the home or in the community	0	1	2	3
11	Meeting the needs of your (other) children	0	1	2	3
12	Meeting your own personal needs	0	1	2	3
13	Meeting the needs of your partner	0	1	2	3
14	Maintaining satisfying friendships for yourself	0	1	2	3
15	Dealing with _____'s sexuality	0	1	2	3
16	Work placements or employment for _____	0	1	2	3
17	Long-term planning for accommodation for _____	0	1	2	3
18	Planning for wills, trusts and guardianships	0	1	2	3
19	Planning for emotional and social support for _____	0	1	2	3
20	Transportation	0	1	2	3
21	Day to day assistance with care of _____	0	1	2	3
22	Time apart from _____	0	1	2	3
23	Dealing with financial issues	0	1	2	3

Section 3: Your Quality of Life

This assessment asks how you feel about different areas of your life.

Please keep in mind your standards, hopes, pleasures and concerns. We ask that you think about your life **in the last two weeks**.

Please read each question, assess your feelings, and circle the number on the scale for each question that gives the best answer for you.

		Very poor	Poor	Neither poor nor good	Good	Very good
1 G1)	How would you rate your quality of life?	1	2	3	4	5

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
2 G4)	How satisfied are you with your health?	1	2	3	4	5

The following questions ask about how much you have experienced certain things in the last two weeks.

		Not at all	A little	A moderate amount	Very much	An extreme amount
3 F1.4)	To what extent do you feel that (physical) pain prevents you from doing what you need to do?	1	2	3	4	5
4 F11.3)	How much do you need any medical treatment to function in your daily life?	1	2	3	4	5
5 (F4.1)	How much do you enjoy life?	1	2	3	4	5
6 F24.2)	To what extent do you feel your life to be meaningful?	1	2	3	4	5

		Not at all	A little	A moderate amount	Very much	Extremely
7 F5.3)	How well are you able to concentrate?	1	2	3	4	5
8 (F16.1)	How safe do you feel in your daily life?	1	2	3	4	5
9 (F22.1)	How healthy is your physical environment?	1	2	3	4	5

The following questions ask about how completely you experience or were able to do certain things in the last two weeks.

		Not at all	A little	Moderately	Mostly	Completely
10 F2.1)	Do you have enough energy for everyday life?	1	2	3	4	5
11 (F7.1)	Are you able to accept your bodily appearance?	1	2	3	4	5
12 F18.1)	Have you enough money to meet your needs?	1	2	3	4	5
13 F20.1)	How available to you is the information that you need in your day-to-day life?	1	2	3	4	5
14 F21.1)	To what extent do you have the opportunity for leisure activities?	1	2	3	4	5

		Very poor	Poor	Neither poor nor good	Good	Very good
15 F9.1)	How well are you able to get around?	1	2	3	4	5

The following questions ask you to say how good or satisfied you have felt about various aspects of your life over the last two weeks.

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
16 F3.3)	How satisfied are you with your sleep?	1	2	3	4	5
17 F10.3)	How satisfied are you with your ability to perform your daily living activities?	1	2	3	4	5
18 F12.4)	How satisfied are you with your capacity for work?	1	2	3	4	5
19 (F6.3)	How satisfied are you with yourself?	1	2	3	4	5
20 F13.3)	How satisfied are you with your personal relationships?	1	2	3	4	5
21 F15.3)	How satisfied are you with your sex life?	1	2	3	4	5
22 F14.4)	How satisfied are you with the support you get from your friends?	1	2	3	4	5

23 F17.3	How satisfied are you with the conditions of your living place?	1	2	3	4	5
24 F19.3	How satisfied are you with your access to health services?	1	2	3	4	5
25 F23.3	How satisfied are you with your transport?	1	2	3	4	5

The following question refers to how often you have felt or experienced certain things in the last two weeks.

		Never	Seldom	Quite often	Very often	Always
26 F8.1)	How often do you have negative feelings such as blue mood, despair, anxiety, depression?	1	2	3	4	5

Do you have any comments about the assessment?

.....
.....
.....
.....

THANK YOU FOR YOUR HELP

2. Family Quality of Life Survey (FQoLS)

Family quality of life

Survey Information and Instructions

Thank you for agreeing to complete this survey.

All the information you give us is confidential. It is important that you answer as many questions as you can, but please feel free to skip any questions that make you feel uncomfortable.

When answering these questions, please think about your experiences **over the last 6 months**. Please use a pencil to shade completely the circles for your answers. If you change any answers, please erase completely any previous answers or any extra pencil marks on the page

Thank you so much for sharing your opinion with us!

We want you to tell us how you feel about your life together as a family.

Your “family” may include many people – mother, father, partners, children, aunts, uncles, grandparents, etc.

For this survey, please consider your family as those people

- Who think of themselves as part of your family (even though they may or may not be related by blood or marriage), and
- Who support and care for each other **on a regular basis**.

For this survey, please **DO NOT** think about relatives (extended family) who are only involved with your family every once in a while. Please think about your family life over the past six months.

Step 1: Importance – First, please shade in the circle in the first set of columns to show how important you think that statement is.

- Shading the **first** circle means you think the statement is only a **little important**.
- Shading the **fifth** circle means you think that statement is **critically important**.

Step 2: Satisfaction – Please shade in the circle in the next set of columns to show how satisfied you are with that statement.

- Shading the **first** circle means you are **very dissatisfied**.
- Shading the **last** circle means you are **very satisfied**.
-

Please remember to answer both **IMPORTANCE** and **SATISFACTION** for each question.

Family Quality of Life.

**For my family to have a good
Life together...**

1. My family enjoys spending time together.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
2. My family members help the children learn to be independent.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
3. My family has the support we need to relieve stress.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
4. My family members have friends or others who provide support.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
5. My family members help the children with schoolwork and activities.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
6. My family members have transportation to get to the places they need to be.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
7. My family members talk openly with each other.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

8. My family members teach the children how to get along with others.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
9. My family members have some time to pursue their own interests.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
10. My family solves problems together.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
11. My family members support each other to accomplish goals.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
12. My family members show that they love and care for each other.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
13. My family has outside help available to us to take care of special needs of all family members.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
14. Adults in my family teach the children to make good decisions.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Family Quality of Life (cont.)

**For my family to have a
good life together...**

15. My family gets medical care when needed.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
16. My family has a way to take care of our expenses.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
17. Adults in my family know other people in the children's lives (friends, teachers, etc.).	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
18. My family is able to handle life's ups and downs.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
19. Adults in my family have time to take care of the individual needs of every child.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
20. My family gets dental care when needed.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
21. My family feels safe at home, work, school, and in our neighborhood.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
22. My family member with additional learning needs has support to accomplish goals at school or workplace.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
23. My family member with additional learning needs has support to accomplish goals at home.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
24. My family member with additional learning needs has support to make friends.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
25. My family has good relationships with the service providers who provide services and support to our family member with additional learning needs.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

General Individual and Family Information

Please answer these questions about **yourself**.

1. What is your gender?

☐ Male

☐ Female

2. What year were you born?

3. What is your relationship to the child or young person with additional learning needs in your family?

☐ Parent (Biological, Step, Foster or Adoptive)

☐ Other relative (grandparent, aunt, uncle, sibling, etc.) Please specify:

☐ Other non-relative (family friend, etc.) Please specify: _____

☐ I am the young person with additional learning needs

3. Childhood behaviour checklist (CBCL)

Below is a list of items that describe some children. For each item that describes your child from the age of 13-16 years please circle 2 if the item was true or often true of your child. If the item was definitely not true of your child please circle 0. Circle 1 if it was sometimes or somewhat true for your child..

	Between 13 years and 16 years		
1. Acts too young for his/her age	0	1	2
2. Allergy (describe)	0	1	2
3. Argues a lot	0	1	2
4. Asthma	0	1	2
5. Behaves like the opposite sex	0	1	2
6. Bowel movements outside the toilet	0	1	2
7. Bragging, boasting	0	1	2
8. Can't concentrate, can't pay attention for too long	0	1	2
9. Can't get his/mind off certain thoughts or obsessions (describe)	0	1	2
10. Can't sit still, restless or hyperactive	0	1	2
11. Clings to adults or too dependent	0	1	2
12. Complains of loneliness	0	1	2
13. Confused or in a fog	0	1	2
14. Cries a lot	0	1	2
15. Cruel to animals	0	1	2
16. Cruelty, bullying or meanness to others	0	1	2
17. Day-dreams or gets lost in his/her thoughts	0	1	2
18. Deliberately harm him/herself or attempts suicide	0	1	2
19. Demands a lot of attention	0	1	2
20. Destroys his/her things	0	1	2
21. Destroys things belonging to his/her family or other children	0	1	2
22. Disobedient at home	0	1	2
23. Disobedient at school	0	1	2
24. Doesn't eat well at home	0	1	2

	Between 13 years and 16 years		
25. Doesn't get on well with other children	0	1	2
26. Doesn't seem to feel guilty after misbehaving	0	1	2
27. Easily jealous	0	1	2
28. Eats or drinks things that are not food (describe)	0	1	2
29. Fears certain animals, situations or places other than school (describe)	0	1	2
30. Fears going to school	0	1	2
31. Fears that he/she might think or do something bad	0	1	2
32. Feels he/she has to be perfect	0	1	2
33. Feels or complains that nobody loves him/her	0	1	2
34. Feels others are out to get him/her	0	1	2
35. Feels worthless or inferior	0	1	2
36. Gets hurt a lot, accident-prone	0	1	2
37. Gets in many fights	0	1	2
38. Gets teased a lot	0	1	2
39. Hangs around with children who get in trouble	0	1	2
40. Hears things that aren't there (describe).....	0	1	2
41. Impulsive or acts without thinking	0	1	2
42. Likes to be alone	0	1	2
43. Lying or cheating	0	1	2
44. Bites fingernails	0	1	2
45. Nervous, highly-strung, or tense	0	1	2
46. Nervous movements or twitching	0	1	2
47. Nightmares	0	1	2
48. Not liked by other children	0	1	2
49. Constipated, doesn't move bowels	0	1	2
50. Too tearful or anxious	0	1	2

51. Feels dizzy	0	1	2
52. Feels too guilty	0	1	2
53. Overeating	0	1	2
54. Overtired	0	1	2
55. Overweight	0	1	2
56. Physical problems with medical cause			
a. Aches or pains	0	1	2
b. Headaches	0	1	2
c. Nausea, feels sick	0	1	2
d. Problems with eyes (describe)	0	1	2
.....			
e. Rashes or other skin conditions	0	1	2
f. Stomach aches or cramps	0	1	2
g. Vomiting, throwing up	0	1	2
h. Other (describe)	0	1	2
57. Physically attacks people	0	1	2
58. Picks nose, skin or other parts of body (describe)	0	1	2
.....			
59. Plays with his/her own sex parts in public	0	1	2
60. Plays with his/her own sex parts too much	0	1	2
61. Poor school work	0	1	2
62. Poorly co-ordinated or clumsy	0	1	2
63. Prefers playing with older children	0	1	2
64. Prefers playing with younger children.....	0	1	2
65. Refuses to talk	0	1	2
66. Repeats certain acts over and over; compulsions (describe).....	0	1	2
67. Runs away from home	0	1	2
68. Screams a lot	0	1	2
69. Secretive, keeps things to him/herself	0	1	2

70. Sees things that aren't there (describe)	0	1	2
.....			
71. Self-conscious or easily embarrassed	0	1	2
72. Sets fires	0	1	2
73. Sexual problems (describe)	0	1	2
.....			
74. Showing off or clowning	0	1	2
75. Shy or timid	0	1	2
76. Sleeps less than most children	0	1	2
77. Sleeps more than most children during the day or night.....	0	1	2
78. Smears or plays with bowel movements	0	1	2
79. Speech problem (describe)	0	1	2
.....			
80. Stares blankly	0	1	2
81. Steals at home	0	1	2
82. Steals outside the home	0	1	2
83. Stores up things he/she doesn't need (describe)	0	1	2
.....			
84. Strange behaviour (describe).....	0	1	2
.....			
85. Strange ideas (describe)	0	1	2
86. Stubborn, sullen or irritable	0	1	2
87. Sudden changes in mood or feelings	0	1	2
88. Sulks a lot	0	1	2
89. Suspicious	0	1	2
90. Swearing or obscene language	0	1	2
91. Talks about killing him/herself	0	1	2
92. Talks or walks in his/her sleep (describe)	0	1	2
93. Talks too much	0	1	2
94. Teases a lot	0	1	2

95. Temper tantrums or a hot temper	0	1	2
96. Thinks about sex too much	0	1	2
97. Threatens people	0	1	2
98. Thumb-sucking	0	1	2
99. Too concerned with neatness or cleanliness	0	1	2
100. Trouble sleeping (describe)	0	1	2
101. Truancy, skips school	0	1	2
102. Underactive, slow moving or lacks energy	0	1	2
103. Unhappy, sad or depressed	0	1	2
104. Unusually loud	0	1	2
105. Uses alcohol, drugs or sniffs glue (describe)	0	1	2
106. Vandalism	0	1	2
107. Wets him/herself during the day	0	1	2
108. Wets the bed	0	1	2
109. Whining	0	1	2
110. Wishes to be of the opposite sex	0	1	2
111. Withdrawn, doesn't get involved with others	0	1	2
112. Worrying	0	1	2
113. Please write in any problem your child has that was not listed above:	0	1	2

PLEASE BE SURE YOU HAVE ANSWERED ALL ITEMS

THANK YOU VERY MUCH FOR YOUR HELP

4.Social Communication Questionnaire (SCQ)

Thank you for taking the time to complete this questionnaire. A few questions ask about several related types of behaviour; please tick yes if any one of these was present. Although you may be uncertain about whether some behaviours were present or not, please do answer 'yes' or 'no' to every question on the basis of what you think.

		YES	NO
1.	Is she/he now able to talk using short phrases or sentences?	<input type="checkbox"/>	<input type="checkbox"/>
	If NO, proceed to question 8		
2.	Can you have a to and fro 'conversation' with her/him that involves taking turns or building on what you have said?	<input type="checkbox"/>	<input type="checkbox"/>
3.	Has she/he ever used odd phrases or said the same thing over and over in almost exactly the same way? That is, either phrases she/he has heard other people use or the ones she/he has made up?	<input type="checkbox"/>	<input type="checkbox"/>
4.	Has she/he ever used socially inappropriate questions or statements? For example, has she/he ever regularly asked personal questions or made personal comments at awkward times?	<input type="checkbox"/>	<input type="checkbox"/>
5.	Has she/he ever got her/his pronouns the wrong way round (i.e. saying 'you' or 'she/he' for 'I')?	<input type="checkbox"/>	<input type="checkbox"/>
6.	Has she/he ever used words that she/he seemed to have invented or made up her/himself, or ever put things in odd, indirect ways, or used metaphorical ways of saying things? For example, saying 'hot rain' for 'steam'?	<input type="checkbox"/>	<input type="checkbox"/>
7.	Has she/he ever said the same thing over and over in exactly the same way, or insisted on you saying the same things over and over again?	<input type="checkbox"/>	<input type="checkbox"/>
8.	Has she/he ever had things that she/he seemed to have to do in a very particular way or order, or rituals that she/he insisted you go through?	<input type="checkbox"/>	<input type="checkbox"/>
9.	Has her/his facial expression usually seemed appropriate to the particular situation, as far as you could tell?	<input type="checkbox"/>	<input type="checkbox"/>

10.	Has she/he ever used your hand like a tool, or as if it were part of her/his own body (e.g. pointing with your finger, putting your hand on a doorknob to get you to open the door)?	<input type="checkbox"/>	<input type="checkbox"/>
11.	Has she/he ever had any interests that preoccupy her/him and might seem odd to other people (eg traffic lights, drainpipes or timetables)?	<input type="checkbox"/>	<input type="checkbox"/>
12.	Has she/he ever seemed to be more interested in parts of a toy or an object (eg spinning the wheels of a car), rather than using the object as it was intended?	<input type="checkbox"/>	<input type="checkbox"/>
13.	Has she/he ever had any special interests that were <u>unusual</u> in their intensity but otherwise appropriate for her/his age and peer group (eg trains, dinosaurs)?	<input type="checkbox"/>	<input type="checkbox"/>
14.	Has she/he ever seemed to be <u>unusually</u> interested in the sight, feel, sound, taste or smell of things or people?	<input type="checkbox"/>	<input type="checkbox"/>
15.	Has she/he ever had any mannerisms or odd ways of moving her/his hands or fingers, such as flapping or moving her/his fingers in front of her/his eyes?	<input type="checkbox"/>	<input type="checkbox"/>
16.	Has she/he ever had any complicated movements of her/his whole body, such as spinning or repeatedly bouncing up and down?	<input type="checkbox"/>	<input type="checkbox"/>
17.	Has she/he ever injured her/himself deliberately, such as by biting her/his arm or banging her/his head?	<input type="checkbox"/>	<input type="checkbox"/>
18.	Has she/he ever had any objects (<u>other</u> than a soft toy or comfort blanket) that she/he <u>had</u> to carry around?	<input type="checkbox"/>	<input type="checkbox"/>
19.	Does she/he have any particular friends or a best friend?	<input type="checkbox"/>	<input type="checkbox"/>

(continued)

For the following behaviours, please focus on the time period between the 4th and 5th birthdays. You may find it easier to remember how things were at that time by focusing on key events, such as starting school, moving house, Christmas time or any events that are particularly memorable for you as a family. If your child is not yet four years old, please consider his or her behaviour in the last 12 months.

20.	When she/he was 4-to-5, did she/he ever talk with you just to be friendly (rather than to get something)?	<input type="checkbox"/>	<input type="checkbox"/>
21.	When she/he was 4-to-5, did she/he ever <u>spontaneously</u> copy you (or other people), or what you were doing (such as hoovering, gardening, mending things)?	<input type="checkbox"/>	<input type="checkbox"/>
22.	When she/he was 4-to-5, did she/he ever spontaneously point at things around her/him just to show you things (not because she/he wanted them)?	<input type="checkbox"/>	<input type="checkbox"/>
23.	When she/he was 4-to-5, did she/he ever use gestures, other than pointing or pulling your hand, to let you know what she/he wanted?	<input type="checkbox"/>	<input type="checkbox"/>
24.	When she/he was 4-to-5, did she/he nod her/his head to mean “yes”?	<input type="checkbox"/>	<input type="checkbox"/>
25.	When she/he was 4-to-5, did she/he shake her/his head to mean “no”?	<input type="checkbox"/>	<input type="checkbox"/>
26.	When she/he was 4-to-5, did she/he usually look at you directly in the face when doing things with you or talking with you?	<input type="checkbox"/>	<input type="checkbox"/>
27.	When she/he was 4-to-5, did she/he smile back if someone smiled at her/him?	<input type="checkbox"/>	<input type="checkbox"/>
28.	When she/he was 4-to-5, did she/he ever show you things that interested her/him to engage your attention?	<input type="checkbox"/>	<input type="checkbox"/>
29.	When she/he was 4-to-5, did she/he ever offer to share things other than food with you?	<input type="checkbox"/>	<input type="checkbox"/>

30.	When she/he was 4-to-5, did she/he ever seem to want you to join in her/his enjoyment of something?	<input type="checkbox"/>	<input type="checkbox"/>
31.	When she/he was 4-to-5, did she/he ever try to comfort you if you were sad or hurt?	<input type="checkbox"/>	<input type="checkbox"/>
32.	When she/he was 4-to-5, when she/he wanted something or wanted help, did she/he look at you and use gestures with sounds or words to get your attention?	<input type="checkbox"/>	<input type="checkbox"/>
33.	When she/he was 4-to-5, did she/he show a normal range of facial expressions?	<input type="checkbox"/>	<input type="checkbox"/>
34.	When she/he was 4-to-5, did she/he ever spontaneously join in and try to copy the actions in social games – such as ‘The Mulberry Bush’ or ‘The Farmer’s in his Den’?	<input type="checkbox"/>	<input type="checkbox"/>
35.	When she/he was 4-to-5, did she/he play any pretend or make-believe games?	<input type="checkbox"/>	<input type="checkbox"/>
36.	When she/he was 4-to-5, did she/he seem interested in other children of approximately the same age whom she/he did not know?	<input type="checkbox"/>	<input type="checkbox"/>
37.	When she/he was 4-to-5, did she/he respond positively when another child approached her/him?	<input type="checkbox"/>	<input type="checkbox"/>
38.	When she/he was 4-to-5, if you came into a room and started talking to her/him without calling her/his name, did she/he usually look up and pay attention to you?	<input type="checkbox"/>	<input type="checkbox"/>
39.	When she/he was 4-to-5, did she/he ever play imaginative games with another child in such a way that you could tell they understood what each other was pretending?	<input type="checkbox"/>	<input type="checkbox"/>
40.	When she/he was 4-to-5, did she/he play co-operatively in games that needed some form of joining in with a group of other children, such as hide and seek or ball games?	<input type="checkbox"/>	<input type="checkbox"/>

Appendix 3

Quantitative phase documentation

- 1. mothers' letter of invitation**
- 2. family members' letter of invitation**
- 3. information sheet**
- 4. consent form**

1. parent letter of invitation

date

Dear

Quality of life in families of young people with additional learning needs

You are invited to take part in the above study in which aims to explore the experiences of families of young people with a range of additional learning needs. This is related to the larger project 'Mental health in young people with additional learning needs' with which you have already helped us.

Taking part will involve completing two short questionnaires and returning them by post. One of the questionnaires can also be completed by other family members over the age of 13, including the young person with additional learning needs. At a later stage, some people may also be invited to take part in an interview with the researcher lasting about one hour. A sheet is enclosed giving more information about the study.

If you would like to take part, or if you would like to ask any questions before deciding, please call me on **0131 537 6257** or complete and return the enclosed consent form in the stamped addressed envelope provided.

Yours sincerely

Vivien Moffat
Research Nurse

2. family member letter of invitation

date

Dear Family Members

Quality of life in families of young people with additional learning needs

You are invited to take part in the above study which aims to explore the experiences of families of young people with a range of additional learning needs. This is related to the larger project 'Mental health in young people with additional learning needs' with which your family has already helped us.

Taking part would involve completing the enclosed questionnaire which would then be returned by post with the questionnaires from other family members. The questionnaire can be completed by any family members over the age of 13. We would ask that each family member completes the questionnaire independently. A sheet is enclosed giving more information about the study.

If you would like to take part, please complete and return the enclosed consent slip with your completed questionnaire. If you would like to ask any questions before deciding, please call me on **0131 537 6257**.

Yours sincerely

Vivien Moffat
Research Nurse

3. Information sheet

Quality of life in families of young people with additional learning needs

You are being invited to take part in this research because you have already helped us with our larger study 'Mental health in young people with additional learning needs'. In that study we recruited almost 500 young people from across Scotland. In most cases we visited parents in their homes to complete the questionnaires about each young person's personality and behaviour. During these meetings many parents told us something of their family situation and the challenges they have faced in relation to the care and education of their son or daughter. We felt that these experiences were important and that many of the things parents told us deserve further investigation. This study has been designed to give parents, families and young people with additional learning needs the opportunity to tell us more about their experiences.

Why has my family been chosen?

We have invited a selection of parents from Edinburgh and Lothian, Lanarkshire and the Glasgow area to take part in this study. We have chosen families to represent a broad cross section of young people with different types and degrees of additional learning needs, including autistic spectrum disorder, as well as a group of families where there is no family member with additional learning needs of any kind. We hope to recruit 100 families.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason. A decision to withdraw, or not to take part, will not affect any help or services you or your child may receive.

What will happen if I take part?

The first stage of the study would involve completing two short questionnaires. We will ask you to return these to us by post, although we would be happy to complete the questionnaires with you if that was preferred. The first questionnaire asks you about the things that matter for your family in order to have a good quality of life. Other members of the family can also fill in this questionnaire if they wish to do so, although we would ask that each family member completes the questionnaire independently. We would give you the number of questionnaires your family requires and a stamped addressed envelope for their return. The second questionnaire can also be returned by post. This questionnaire asks you how stressful certain issues are for you and your family. The questionnaires are about issues such as your child's education, any diagnosis he or she may have, life within your community for your child and your family and the amount of help or support you and your family receive.

Some people will be invited to take part in the second stage of the study. This would involve an interview with the researcher lasting about one hour. If you were willing, we would like to audio tape the interview. During the interview you would be asked a bit more about the issues you told us were most stressful for you in your questionnaire replies.

In particular you would be asked:

- What challenges have you faced and how have you coped?
- What successes have you achieved?
- How have things changed over time?
- What would you like to see changed that would help you and other parents in the future?

Will my taking part in this study be kept confidential?

Any information you give will be completely confidential and neither you nor your child would be identifiable in any of the results of the study.

Who is organising and funding the research?

The study is being carried out by staff from the Division of Psychiatry at the University of Edinburgh and is funded by the Medical Research Council. The study is being done, in part, to contribute to a PhD by Vivien Moffat.

We greatly appreciate your help with this study and would not want you to be out of pocket. Any expenses such as travel or subsistence should you choose to complete the questionnaires or interview at the University or loss of earnings should you miss time at work in order to take part will be met by the Medical Research Council.

If you have any questions about the study or would like to discuss it further before deciding whether to take part, please contact Vivien Moffat on 0131 537 6257. If you would like to speak to someone who is not directly involved in the study and could give you impartial advice, please call Professor David Cunningham Owens on 0131 537 6262.

4. consent form

Quality of Life in Families of Young People with Additional Learning Needs

Please tick box

- | | |
|--|--------------------------|
| I consent to completing questionnaires related to the above study | <input type="checkbox"/> |
| I have read and understand the information sheet | <input type="checkbox"/> |
| I understand that I am not obliged to take part and am free to withdraw from the study at any stage | <input type="checkbox"/> |
| I understand that this is non-therapeutic research from which I cannot expect to derive any personal benefit | <input type="checkbox"/> |

Signature.....Date.....

Please print name.....

Address.....

.....

Post code

Telephone.....

When is a good time to contact you?.....

Appendix 4

Qualitative phase documentation

- 1. letter of invitation for interview**
- 2. consent form**
- 3. FSCI interview questions**

1. letter of invitation for interview

Date

Dear

Quality of life in families of young people with additional learning needs

Thank you for helping with the above study by returning the two questionnaires. You may remember that one of the questionnaires asked you about how stressful certain issues were for you. You are now invited to take part in an interview in which you will be asked a bit more about the issues that you identified as being most stressful.

The interview would last about one hour and could be done at your home at a time that suits you. If you would prefer to come here to the University, we would be happy to arrange this and to cover any travel or other expenses involved. If you agree, we would like to audio tape the interview.

If you would like to take part, or if you would like to ask any questions before deciding, please call me on **0131 537 6257** or complete and return the enclosed consent form in the stamped addressed envelope provided.

Yours sincerely

Vivien Moffat
Research Nurse

2. consent form for interview

Quality of Life in Families of Young People with Additional Learning Needs

Please tick box

I consent to taking part in an interview in relation to the above study

☐

I give permission for the interview to be audio-taped

☐

I understand that I am not obliged to take part and am free to withdraw from the study at any stage

☐

I understand that this is non-therapeutic research from which I cannot expect to derive any personal benefit

☐

Signature.....Date.....

Please print name.....

Address.....

.....

Post code

Telephone.....

When is a good time to contact you?.....

3.FSCI interview questions

- a) What challenges have you faced in this particular issue?

- b) What successes have you had and how did you achieve them?

- c) How have you tried to cope with this challenge
Why did you choose this strategy?
Has this been helpful?

- d) Has the stress related to this issues changed:
I) over the last 5 years?
II) in the last year?

If changes in stress have occurred, why?

- e) What would you like to see change that would help make this experience easier for you and for parents of other children with special needs?

Appendix 5

Qualitative analysis code lists

- 1. Alphabetical list of codes**
- 2. List of categories with codes assigned**
- 3. Categories in relation to themes**

1. Alphabetical list of codes

acceptance	making friends
anger	making sense
aspiration	MMR
becoming an adult	mother's instinct
better off than others	music
breakdown in relationship with service providers	my child is important
bullying	no time for self
coping	normal teenage behaviour
diagnosis	not being believed
disabled identity	not knowing
doing my best	optimism
effect on sib	others dont understand
embarrassment	outlook changing
emotional support	over-involvement
family	parents' role
family relationships	parents' fault
fear of vulnerability	personal growth
fears for future	positive attitude
feel sorry for	positive experiences
form-filling	positive self image
funding	practical support
gap after school	problem solving
guilt	professional support
hard for service providers	raising awareness
having to explain	sadness
having to fight	same as everybody else
helping others	school
informal info sources	self awareness
isolation	self doubt
jobs/career plans	shared experience
just bad	social acceptance
lack of confidence in services	someone to talk to
lack of independence	stigma
lack of info	stress changing
lack of opportunity	support needs
lack of provision	take it as it comes
learning	things that work
letting go	untapped potential
limitations	waiting
looks 'normal'	what might help
	what s/he wants

2. List of categories with codes assigned

Emotion focused coping

anger
embarrassment
emotional support
feel sorry for
guilt
self doubt
sadness

Problem solving

doing my best
having to fight
helping others
learning
letting go
things that work

Positive reappraisal

better off than others
making sense
my child is important
optimism
personal growth
positive attitude
positive experiences

Family relationships

effect on siblings
family relationships
having to explain
others don't understand
parents' fault
support needs

Positive outcomes and attitudes

aspiration
family relationships
helping others
learning
letting go
mother's instinct
outlook changing
personal growth
positive attitude
positive experiences
positive self image
raising awareness
things that work
untapped potential

Transition

becoming an adult
fears for future
form-filling
gap after school
jobs/career plans
lack of independence
lack of opportunity
letting go
normal teenage behaviour
school
stress changing
untapped potential
what s/he wants

Relationships with service providers

breakdown in relationship with service providers
form-filling
hard for service providers
having to fight
lack of confidence in services
not being believed
others don't understand
professional support
school
someone to talk to
things that work
waiting
what might help

Young people-self image

becoming an adult
bullying
disabled identity
isolation
jobs/career plans
positive self image
same as everybody else
self awareness
social acceptance
stigma
what s/he wants

Effect of ASD diagnosis

diagnosis
disabled identity
fear of vulnerability
fears for future
feel sorry for
form-filling
making sense
mother's instinct
others don't understand
outlook changing
practical support
professional support
raising awareness
shared experience
someone to talk to
stigma
stress changing

Uncertainty

fears for future
gap after school
jobs/career plans
lack of confidence in services
lack of info
making sense
MMR
not knowing
someone to talk to
waiting

Hopes for future

aspiration
becoming an adult
jobs/career plans
making friends
optimism
outlook changing
positive experiences
same as everybody else
social acceptance
things that work
what s/he wants
what might help
untapped potential

Changes wanted

jobs/career plans
professional support

raising awareness
same as everybody else
school
social acceptance
things that work
what might help
what s/he wants

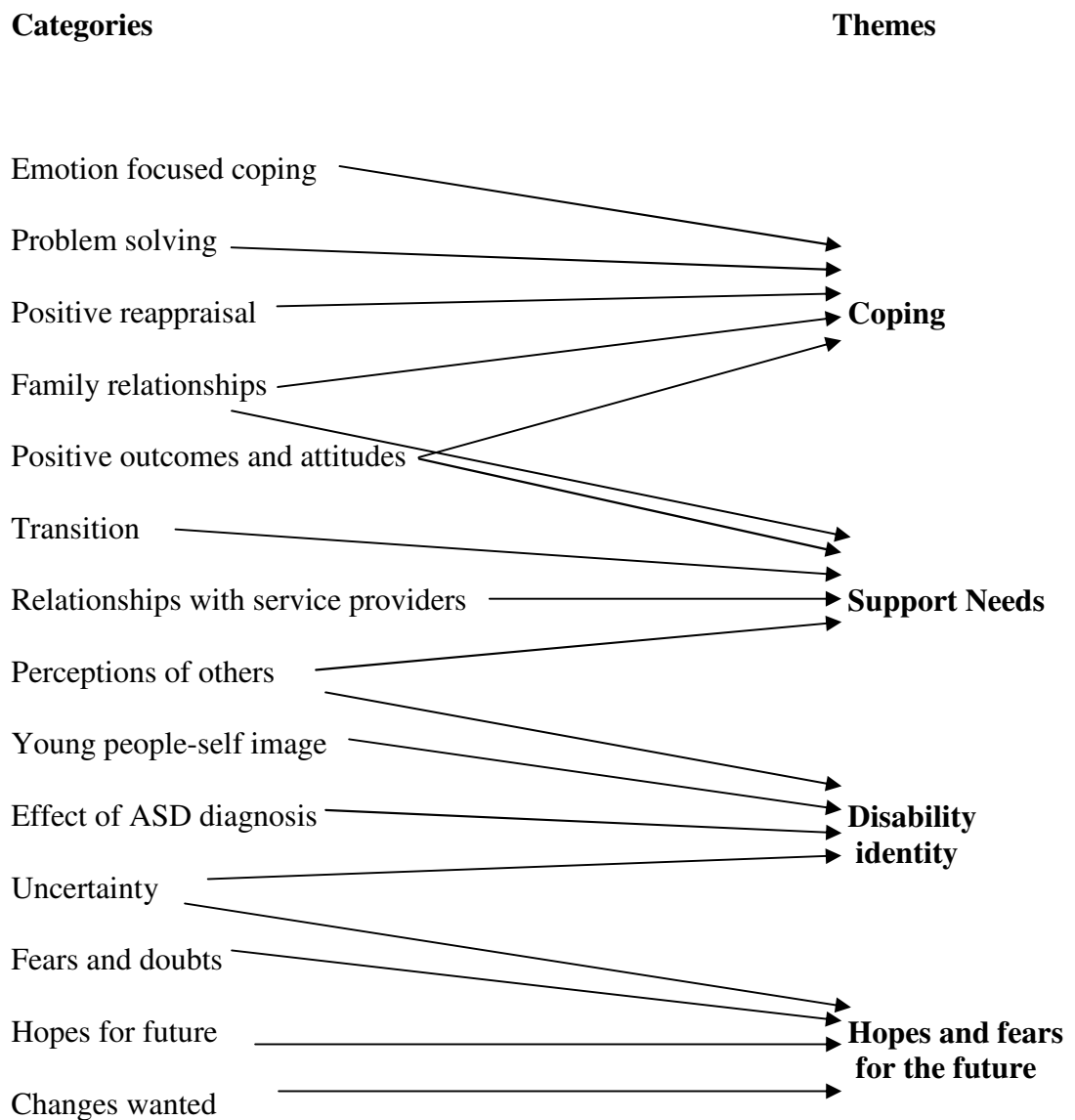
Fears and doubts

fear of vulnerability
fears for future
feel sorry for
funding
isolation
jobs/career plans
just bad
lack of confidence in services
lack of info
lack of provision
lack of opportunity
limitations
not knowing
others don't understand
sadness
school
self doubt
social acceptance
stigma
stress changing
what s/he wants
waiting
support needs

Perceptions of others

embarrassment
family
having to explain
just bad
looks 'normal'
not being believed
others don't understand
outlook changing
parents' fault
raising awareness
same as everybody else
what might help
stigma
social acceptance

3.Categories in relation to themes



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